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Splenogonadal fusion: A case of two lesions

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A B S T R A C T

Splenogonadal fusion is a rare congenital anomaly that presents as a testicular mass and is clinically difficult to distinguish from primary malignancy. It is a benign condition which commonly results in a radical orchidectomy. This case report describes a 22-year-old male with a new multi-focal lesion discovered on self-examination. It aims to provide additional information in hopes of supporting other surgeons to diagnose this condition and prevent unnecessary radical orchidectomy.

Introduction

Solid intratesticular masses in young men are considered malignant until proven otherwise. Appropriate work-up includes clinical history and examination, ultrasound (USS) imaging, and serum tumor markers. When these investigations return reassuring, management of a presumed benign testicular mass can be challenging. The case below details this experience and hopes to provide further evidence to reduce the chance of an unnecessary radical orchidectomy being performed for a benign testicular lesion.

Case presentation

A 22-year-old male was seen in the Urology outpatient clinic after noticing a hard mass within the upper pole of his left testicle on self-examination. His background was significant for an epididymal lump on the same side presumed to be an epididymal cyst. Otherwise he was fit and well with no remarkable past medical history. He described the mass as increasing in size over the past three months with associated lower abdominal pain which radiated into his left testicle. On examination, there were two discrete palpable masses, one near the epididymis and the other unable to be differentiated from the testicle itself. A scrotal USS showed a solid, hypoechoic mass within the upper pole of the testicle measuring 9 × 8 × 5 mm (Fig. 1). The mass was hypervascular with significant internal vascularity. Additionally, there was a second mass medial and separate to the epididymal head measuring 7 × 6 × 7 mm with a similar hypervascular appearance. There were no enlarged inguinal or para-aortic nodes seen on imaging and his chest x-ray was unremarkable. Serum tumor markers including alpha-fetoprotein (AFP), beta human chorionic gonadotropin (β-hCG), and lactate dehydrogenase (LDH) were normal. A repeat USS was performed which showed the lesions did not increase in size over a 2 month period.

His case was discussed at multiple multidisciplinary meetings with the consensus for continued USS surveillance on a 6-monthly basis. However due to the uncertainty regarding these lesions and continued lower abdominal pain, the patient opted for a radical orchidectomy. Intraoperative appearances of the testicle revealed two areas of macroscopic difference. The first lesion was firm, brown, and outside of the testicle parenchyma. Additionally, it was encapsulated by a thin white fibrous tissue. The second lesion was similar in appearance but was within the testicular parenchyma and not encapsulated. The histology results showed two foci of ectopic splenic tissue comprising of well demarcated, encapsulated nodules of red and white pulp (Fig. 2). One of the nodules was attached to the tunica albuginea and lied outside the testis (Fig. 3-A), while the other nodule lied within the testicular parenchyma surrounded by normal seminiferous tubules (Fig. 3-B). The features were consistent with Splenogonadal fusion, discontinuous type.

Discussion

Splenogonadal Fusion (SGF) is a rare congenital anomaly with only approximately 200 cases reported since first being described by Bostroem in 1883.[1,2] It occurs between the 5th and 8th weeks of gestation as the embryonic gut rotates bringing splenic tissue in apposition with one of the urogenital folds.[2] Ninety-eight percent of cases previously described are on the left hand side.[3] Commonly it has been identified at autopsy, however in vivo it mostly presents as either a scrotal mass, inguinal hernia, or cryptorchidism.[2] The latter presentation is responsible for its association with primary malignancy, even though it is rare.[2] Reports in the literature also describe an association with congenital anomalies including limb defects, cleft palate, anal atresia, diaphragmatic hernia, and cardiac malformations.[1–4] In a

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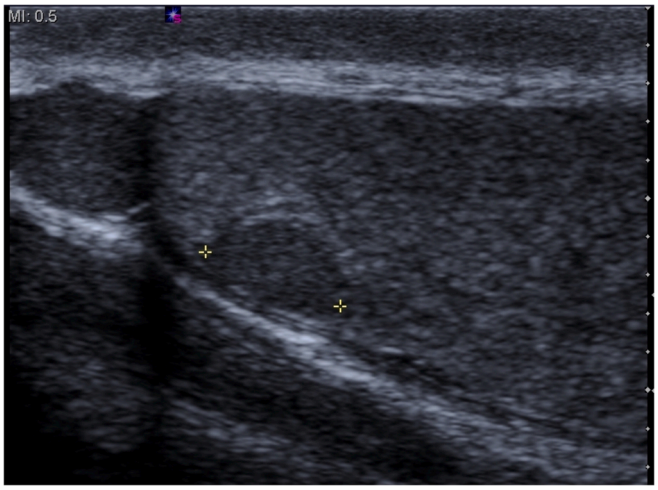

Fig. 1. Longitudinal ultrasound image of the left testicle demonstrating a solid, hypoechoic mass within the upper pole.

review of previous case reports, congenital anomalies were seen in 20–48% of individuals.[2,4,5] The two types of SGF which have been described in the literature are continuous and discontinuous.[1–3] The more prevalent continuous type is characterized by a persisting fibrous tissue connection between the spleen and the gonad. This connection does not exist in the discontinuous type, which is rarer accounting for 44% of all reported cases.[4]

Seventy-two percent of SGF cases have occurred in individuals less than 20 years of age.[4] Painless testicular masses in this age range should be treated as malignancy until proven otherwise. The importance of not missing malignancy combined with not readily available diagnostic testing helps explain why SGF has resulted in an unnecessary radical orchidectomy in 37% of cases.[2] Radiocolloid spleen scintigraphy has been used to identify accessory splenic tissue using Technetium-99 m, however at smaller centers this technology may not be readily available.[5] Additionally, if the surgeon is confident the mass is benign an intraoperative frozen section can be taken to confirm the diagnosis, and preserve the remaining testicular tissue.[5] Though complicating factors is the possibility of concurrent malignancy with SGF.[5] This risk was particularly important with regards to our patient as they had two separate lesions. No previous literature has described discontinuous type SGF with two discrete lesions within one testicle.

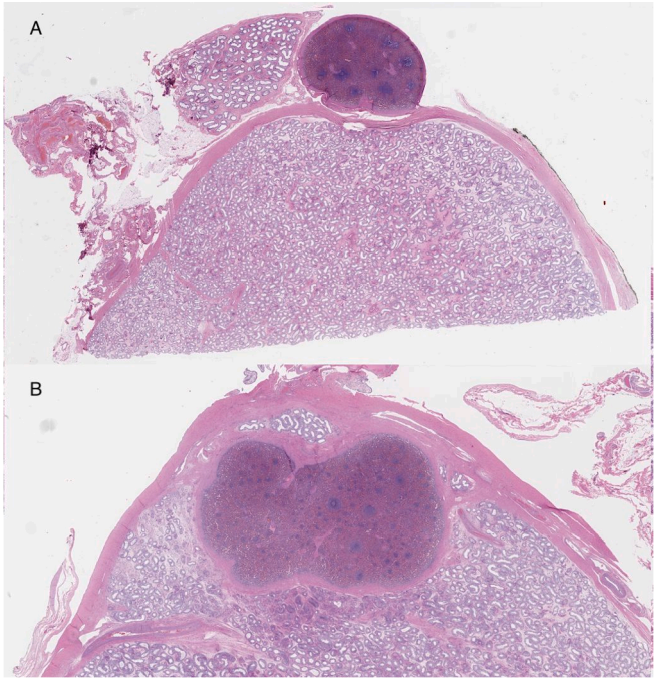

Fig. 3. Microscopic appearances of left testicle demonstrating: (A) Ectopic Splenic tissue located outside the testicular parenchyma, attached to tunica albuginea. (B) Ectopic splenic tissue lying within the testicular parenchyma.

Conclusion

The diagnosis of SGF is tricky and often surgeons opt for radical management to minimize the risk of a missed malignant process. Radiocolloid imaging has been used to diagnose SGF pre-operatively but the surgeon must have a high pre-test probability and it is not 100% sensitive or specific. Additional literature describing the course and progression of SGF will allow surgeons to be more confident in their diagnosis and monitoring management plan. This case report

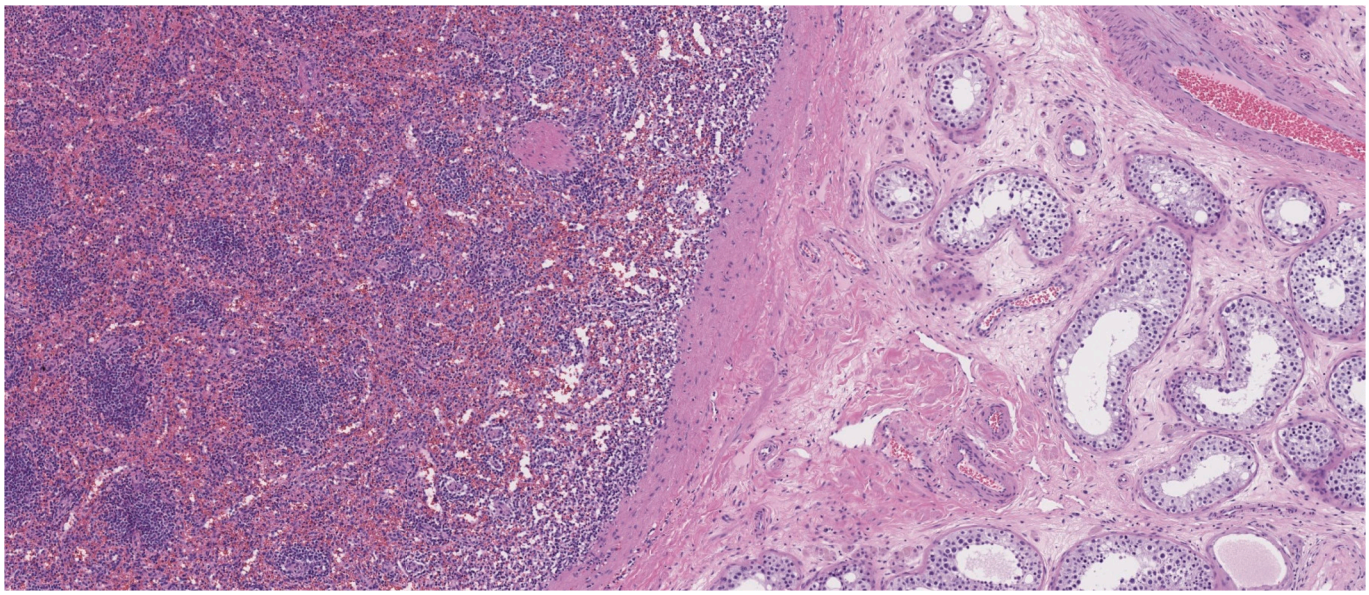

Fig. 2. Microscopic appearances showing well demarcated, encapsulated red/white pulp next to seminiferous tubules. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

introduces new characteristics (i.e. two lesions) and provides further information on the course of SGF in an attempt to reduce the rate of unnecessary radical orchidectomy.

Consent

The authors have received written consent from the patient to perform this case report.

Conflict of interest

The authors declare that they have no potential conflict of interest.

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