Splenogonadal fusion: Report of two cases and literature review

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OBJECTIVE: To suggest a reasonable consideration to avoid the maltreatment of splenogonadal fusion.

METHODS: A 6-year-old male patient (Case 1) underwent unilateral orchiectomy for a suspected tumor of the testis. A 12-year-old male patient (Case 2) underwent bilateral groin exploration and urethroplasty. Postoperative abdominal magnetic resonance imaging revealed an oval-shaped mass in the lower pole of the left kidney and no abnormalities in the spleen. Laparoscopy was considered.

RESULTS: In Case 1, a red mass was found in the upper pole of the testis; the mass occupied one-third of the testis volume. No connection with the cord was found. In Case 2, a red testis-like mass in the lower pole of the left kidney was found and resected. Orchidopexy was performed for the right testis. Postoperative pathology confirmed splenogonadal fusion.

CONCLUSION: Splenogonadal fusion is a rare congenital abnormality. Most patients undergo unnecessary orchiectomy because of suspicion of a primary testicular neoplasm. Common imaging examinations, negative serologic examinations, and 99mTc-sulphur colloid liver-spleen scans can help to achieve a diagnosis. Careful review of the chronicity of the lesion may suggest a benign growth.

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1. Case reports

1.1. Case 1

A 6-year-old male patient was admitted in April 2002 for treatment of a painless mass in the left testicle. It was seen at birth and grew slightly with age. On physical examination, the left scrotum was slightly flabby compared with the right. There was no inflammation, but a swollen testicle was felt in the scrotum (approximately 2.5 × 2.5 × 4.5 cm). It was fairly firm and painless. Testicular carcinoma was suspected and surgery was performed via a groin incision. A reddish brown mass was attached to the upper pole of the left testis. The mass was confined within an intact capsule and occupied one-third of the testis volume. Postoperative microscopic examination revealed splenic tissue that was separated from the surrounding compressed testicular tissue.
doubtful tumor case was removed. Pathological examination of the left specimen revealed splenogonadal fusion (Fig. 1).

2. Discussion

Splenogonadal fusion is a rare congenital anomaly and is most common in children and adolescents [1,2]. Splenogonadal fusion almost always occurs on the left side and is more common in males than females (15:1–20:1) [1,3]. The male preponderance may be because the male sex gland is superficially located and easily discovered. The two above-mentioned cases were both male, and both occurred on the left side.

Splenogonadal fusion can be classified into two types, continuous and discontinuous, but there are no significant clinical differences between the two. Le Roux and Heddle [4] speculated that discontinuous splenogonadal fusion is a rare type of lienculus. The two above-mentioned patients had discontinuous splenogonadal fusion. Cryptorchidism and inguinal hernia are the most common malformations associated with splenogonadal fusion. Approximately 31% of patients have cryptorchidism or inguinal hernia, and 59% of the cryptorchidism is bilateral [5]. Case 1 in this report had no associated malformation. Case 2 had bilateral cryptorchidism and hypospadias.

Splenogonadal fusion typically presents as an asymptomatic testicular mass [6]. Other manifestations include acute testicular pain and swelling caused by ectopic splenic tissue infections [7]. Splenogonadal fusion is difficult to diagnose before surgery because it lacks characteristic features of the disease. Case 1 was under treatment because of a left scrotal mass. Case 2 was under treatment for bilateral cryptorchidism. Neither case was accurately diagnosed. The lack of awareness of this disease is a major factor in misdiagnosis. Imaging can help with diagnosis (e.g., B-type ultrasonography, computed tomography, MRI, and 99TcM spleen scanning) [6], and laparoscopy is especially suitable for diagnosing and treating high cryptorchidism patients [8].

Once an accurate diagnosis is achieved, surgery is not needed in the absence of significant complications, especially in the scrotum. Careful observation is recommended in these cases. Even if surgery is performed, the testes can be preserved in most cases. Splenic tissue can be easily separated from the gonad; as a result, the testes should be retained as much as possible, unless they cannot be moved down into the scrotum in cases of that both vessels and vas deferens are the shortest in the high cryptorchidism. Part of the high cryptorchidism patients whose vas deferens is long enough could be adopt to Fowler-Stephen type orchidopexy. Pay attention to the risks that associated with an orchidopexy in a 12 year old male would be reduced fertility, infertility, and cryptorchidism malignant transformation. When a tumor is suspected, frozen section analysis can help with diagnosis. No malignancy of splenogonadal fusion has yet been reported.

In conclusion, the primary aim of diagnosis is to rule out malignancy. The following three steps should be taken for diagnosis and treatment. First, a benign condition should probably be considered when a mass is found at birth and then grows slowly for several years. Second, various imaging (e.g., B-type ultrasonography, computed tomography, MRI, and 99TcM spleen scanning) studies should be performed to investigate the nature of the mass. There also should be blood work to help with diagnosis. Third, in doubtful cases, a surgical biopsy should be performed, preferably before an incision is made (e.g., needle biopsy, punch biopsy, or classical bivalve biopsy of a regional node) for strongly doubtful tumor case of the high cryptorchidism. If malignancy is proven, radical resection should be immediately performed. For unproven malignancy, orchietomy is adequate for further pathological study. In cases proven benign but in which the organ has been opened, splenic tissue can be removed. Surgical excision may not be needed for splenogonadal fusion.

References