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Case Report

Leiomyoma of Testis: A Case Report and Literature Review

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Abstract ***Leiomyoma* is a benign smooth muscle neoplasm that is very rarely (0.1%) premalignant. They can occur in any organ, but they are infrequently found in the genitourinary tract. In the genitourinary tract, the renal capsule is the most common site of involvement. Intrascrotal leiomyomas are infrequently seen. Leiomyoma of testis is extremely rare. Herein, we report a case of leiomyoma of the testis.**

Keywords: **testis, leiomyoma**

Introduction

Leiomyoma is a benign neoplasm that may arise from any structure or organ containing smooth muscles. In the genitourinary tract, the renal capsule capsule is most commonly involved[1], but this tumor has also been reported in the epididymis , spermatic cord, and tunica albuginea. Intratesticular leiomyoma is extremely rare. Nino-Murcia and Kosek reported the first case of an intratesticular leiomyoma in 1989[2]. Herein, we report a case of leiomyoma of the testis.

Case Report

A 43-year old male patient came to our hospital with chief complaint of right-sided testicular swelling for 17 years, gradually increasing in size with mild right lower abdominal pain 1 year prior to admission. On physical examination, we found an enlarged, hard consistency mass at the lower part of the right testis with mild tenderness at the right epididymis. The right spermatic cord was unremarkable. The left testis, epididymis, and spermatic cord were normal. Scrotal ultrasonography showed a normal right testis located at the upper part of the scrotum with minimal distorted orientation, measuring 3.2 × 1.9 × 2.8 cm in size. The right epididymis was not visualized. There was a well-defined heterogeneous mass with internal vascularity in right scrotum, possibly arising from epididymis or spermatic cord. The left testis was also normal, measuring 2.0×2.7×2.3 cm in size. The left epididymal head was unremarkable. Minimal free fluid was seen in both scrotal sacs (Figure 1).

A tumor marker study revealed that α-fetoprotein (AFP), level was 0.8 IU/mL and the β-human chorionic gonadotropin (β-HCG) level was 0.10 mIU/mL, both of which were within normal limits. The patient underwent a right inguinal exploration and a radical orchiectomy. Operative findings were that of a well demarcated mass measuring 5.0 × 3.5 × 3.0 cm at the peritesticular capsule. The cut surface of the mass

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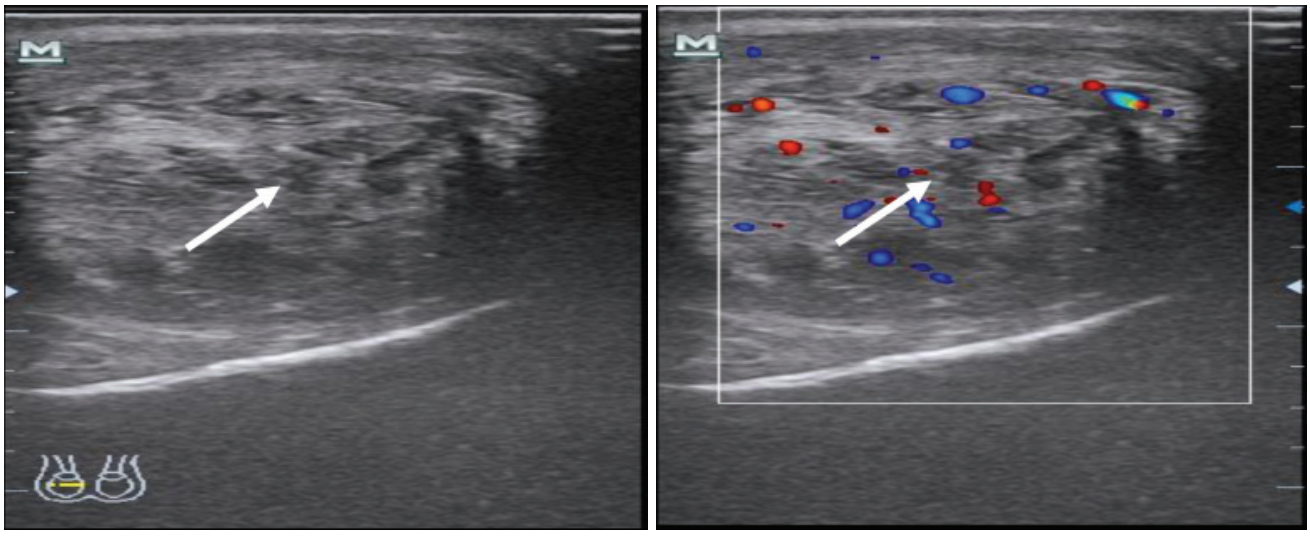

Figure 1 Ultrasound of the right testis showing a well-defined heterogeneous mass with internal vascularity in the right scrotum (white arrows).

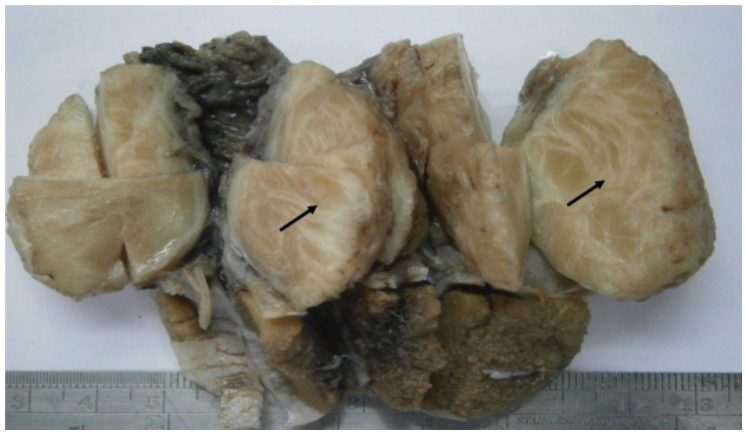

Figure 2 Gross examination of the right testis showing a well demarcated mass at the peritesticular capsule and the gray white, whorl-like appearance of the cut surface (black arrows). The testicular parenchyma, tunica albuginea, and epididymis were normal.

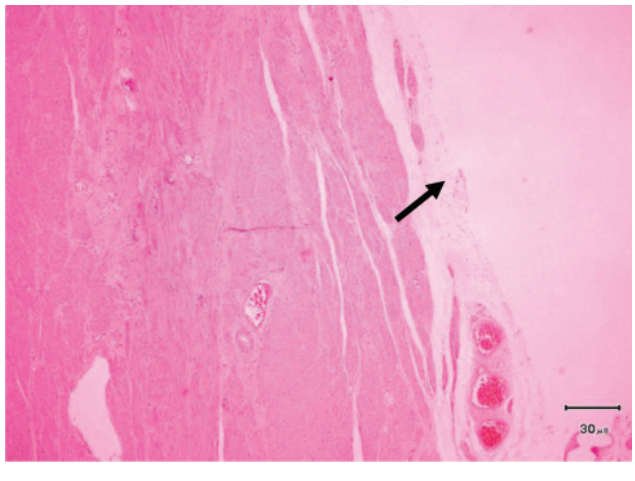

Figure 3 Histological examination of the right intratesticular leiomyoma showing an encapsulated tumor comprising of interlacing fasicles of smooth muscle cells without significant nuclear atypia (black arrows).

showed gray-white, whorl-like appearance. The tunica albuginea was intact and smooth. The rest of the testicular parenchyma was unremarkable. The right epididymis could be identified (Figure 2). Histological examination showed encapsulated tumor comprising interlacing fascicles of smooth muscle cells without significant nuclear atypia. Neither necrosis nor mitotic figures were found. The right epididymis was not remarkable. The spermatic cord margin was free from the tumor (Figure 3). The pathological diagnosis was leiomyoma of the right testis. The postoperative course was uneventful.

Discussion

Albert and Mininberg reported the first case of testis-associated leiomyoma in 1972[3]. We reviewed the literature and found 19 cases previously reported. Chen et al. in 2007 reviewed the literature and found 17 cases, and also proposed using "testis-associated leiomyoma" to describe these lesions. They found that 10 of 17 cases did not involve the testicular parenchyma, while in 7 cases the tumor arose within and displaced part of testicular parenchyma. The mean age (standard deviation) of patients was 52.1 (18.9) years (range, 10-85 years). Six patients (35%) had left testicular involvement, eight patients (47%) had a right side mass, and three patients (18%) had bilateral tumors. In 16 of 17 cases, patients reported slowly growing, painless intrascrotal masses, and only 1 patient suffered from a tender mass due to acute torsion[4].

In 2011 Kullolli et al. reported the 18th case of leiomyoma of testis, found on the left side[5], and in 2012 Bremmer et al. reported the 19th case of leiomyoma of tunica albuginea on the right side of the scrotal sac[6].

We report a case of intratesticular leiomyoma, involving the lower part of the right testis. The duration of symptoms was 17 years. The tumor gradually increased in size, was only mildly tender, and had a hard consistency. Sonography is the imaging modality of choice for evaluating intrascrotal pathology. The sonographic features of leiomyomas included solid hypoechoic or heterogeneous masses that may or may not contain shadowing calcification[7]. In our patient ultrasonography showed a well-defined heterogeneous mass with internal vascularity.

The origin of the intratesticular leiomyoma is controversial, but more recently it is thought to arise from the contractile cells in the tunica propia of seminiferous tubules[8]. Histologically, the characteristic features include the presence of elongated spindle shaped cells with eosinophilic cytoplasm. The nuclei can be normomorphic or oval in shape and usually seen at the center of the cell. The cells tend to be packed and overlapping and can be arranged in intertwining fasciculi[9]. In our patient, histological examination showed an encapsulated tumor comprising interlacing fascicles of smooth muscle cells without significant nuclear atypia. Neither necrosis nor mitotic figures were found.

Chiong et al. in 2004 suggested that inguinal exploration of suspicious scrotal masses is mandatory[10]. They recommend routine intraoperative frozen section biopsy as it may allow for testicular preservation. In our case we undertook a right inguinal exploration and a radical orchiectomy, because we could not distinguish clinically between benign lesions and the more common testicular cancer, and frozen section biopsy was not available at our institute. However serum tumor markers including AFP and β-HCG might help in distinguishing between benign and malignant testicular mass. In our case, AFP and β-HCG levels were within normal limits.

Conclusions

Testicular leiomyomas are a benign and very rare neoplasm. This tumor is generally slow-growing and asymptomatic. Serum tumor marker levels including AFP and β-HCG are usually within normal limits. Ultrasonography is the imaging modality of choice for evaluating intrascrotal pathology. Intrascrotal leiomyoma is indistinguishable from a malignant testicular neoplasm, but is similar in appearance to other leiomyomas occurring elsewhere in the body. Despite its benign nature, radical orchiectomy remains the treatment of choice, because the lesion cannot be distinguished clinically from the more common testicular cancers. A preoperative needle biopsy or intraoperative frozen section biopsy may allow for testicular preservation.

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