Int J Clin Exp Pathol 2015;8(8):9703-9705
www.ijcep.com /ISSN:1936-2625/IJCEP0010986

Case Report

Leiomyomas of the bilateral tunica albuginea of testes: a case report

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Received June 2, 2015; Accepted July 21, 2015; Epub August 1, 2015; Published August 15, 2015

Abstract: Leiomyoma of the bilateral testicular tunica albuginea is extremely rare. To our knowledge, there are only 3 definitely reported cases. This is the first report of bilateral testicular tunica albuginea leiomyomas as a potential cause of male infertility. Herein, we report a case of a 47-year-old man who presented with painless bilateral testicular masses for more than 30 years, besides he also suffered from unexplained infertility. The complete resection of the tumors was performed. The final pathological diagnosis was leiomyomas of the bilateral tunica albuginea. Postoperatively, the patient underwent testicular biopsy. Histopathology confirmed moderate atrophy of bilateral testes, and the number of spermatogenic cells in the seminiferous tubules were significantly decreased. In this case, bilateral testicular dysplasia is the root reason for the patient's infertility. Thus, despite the benign nature of bilateral testicular tunica albuginea leiomyomas, they may cause bilateral testicular hypoplasia and infertility in men. In the case of men with fertility requirements, early local mass excision is often necessary.

Keywords: Leiomyoma, tunica albuginea, testicular dysgenesis, infertility

Introduction

In the male genitourinary tract, the most common location for leiomyoma is the renal capsule [1]. Intrascrotal leiomyomas are uncommon, but they have been found in many locations, including the epididymis, spermatic cord, tunica dartos, tunica albuginea, and body of the testis [2]. Leiomyoma of the tunica albuginea is extremely rare. This is the first report on a case of bilateral testicular tunica albuginea leiomyomas as a potential cause of male infertility.

Case report

A 47-year-old man complaining of a firm, painless mass in bilateral testes that had been present for more than 30 years was presented to the urological department in our hospital, concurrently he was suffering from unexplained infertility. The masses progressively grew in size, particularly in the left. The patient had no remarkable previous medical history.

The physical examination disclosed hard, smooth and non-transilluminating masses without obvious adhesion to the surrounding tissues in the lower poles of the bilateral testes. The bilateral testes appeared to be very small. Inguinal lymph nodes were not touched. Blood biochemistry and urinalysis results were within normal limits. Testicular tumor markers studies, including alpha-fetoprotein and beta human chorionic gonadotropin, showed no pathologic results.

Testicular ultrasound assessment results (**Figure 1**) indicated a hypoechoic heterogeneous tumor measuring 26 mm × 22 mm within the lower pole of the left testis, with normal blood flow signals inside the mass. At the lower pole of the right testis, a much smaller mass (14 mm × 11 mm) with a similar sonographic appearance and blood flow signals was detected. The bilateral testes were smaller than normal. Bilateral inguinal exploration found two well-circumscribed tumors in the periphery of the bilateral testes abutting the tunica albuginea and bilateral testicular dysgenesis obviously. The complete resection of the tumors was accomplished without harming the testes and epididymides. The patient's convalescence was uneventful.

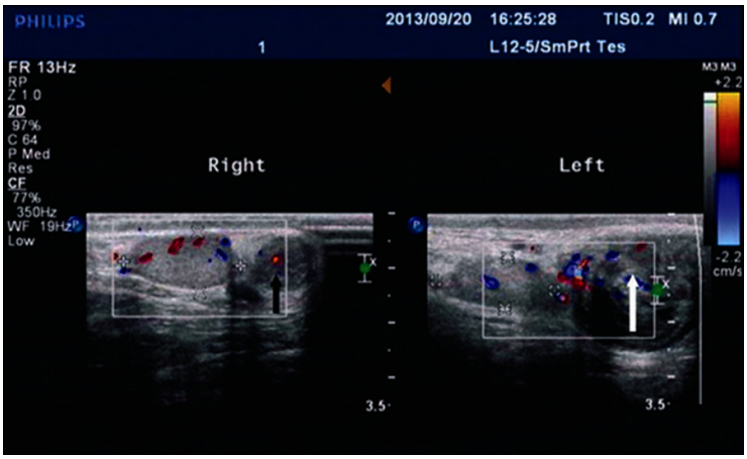

Figure 1. Sonography showed a tumor of 1.4 cm × 1.1 cm in size (black arrow) beside the small hypoplastic right testis, and a tumor of 2.6 cm × 2.2 cm in size (white arrow) beside the small hypoplastic left testis.

Gross pathological examination revealed two discrete nodular masses with complete fibrous capsule. The cut surface of the tumors showed a well-defined gray-white, firm, whorled appearance. Microscopic evaluation showed that the tumors were composed of interlacing uniform spindle cells with blunt-ended elongated nuclei and eosinophilic cytoplasm (**Figure 2**). No evidence of cellular atypia or mitosis was observed. Immunohistochemical staining was performed. The tumor cells were diffusely positive for SMA and desmin, and negative for S100 and myoglobin. Ki-67 showed low focal positivity (< 5% of tumor cells).

Two months postoperatively, our patient underwent a series of fertility examinations, including chromosome, serum levels of six steroid sex hormones, semen and prostatic fluid analysis and so on. Except for low sperm counts, poor sperm motility and viability, the other parameters were normal. Subsequently, the patient underwent testicular biopsy. Histopathology confirmed moderate atrophy of bilateral testes (**Figure 3**).

Discussion

Leiomyoma of the testicular tunica albuginea is rare. Albert and Mininberg described the first case in 1972 [3]. An extensive literature review reveals that only six cases have been reported thus far. This tumor can affect all age groups, but the most common time of presentation is during the sixth decade of life. As of this writing, only three cases of bilateral leiomyomas have been reported [4-6]. The tumor slowly increases in size and has a long history of painless mass. Leiomyoma is a benign tumor, and neither invasion nor metastasis has been observed thus far [6].

At present, surgical local resection is still the main method for the treatment of leiomyoma of the testicular tunica albuginea. Radical orchiectomy is often unnecessary, except when the tumor adheres closely to the adjacent testis or is regarded as malignant. Frozen section examination for intraoperative diagnosis should be performed. In our patient, the tumors were easily separated from the testes and epididymides. Thus, only local masses excision with inguinal incision was performed. The other thief complaint was infertility for our patient. Postoperatively, a series of fertility examinations showed only low sperm counts, poor sperm motility and viability. In this case, the testicular tissues had been oppressed by the surrounding tumors for more than 30 years, during which the testes were in the stage of development. We speculated that the long-term oppression led to testicular dysplasia, which may be one of the root reasons for the patient's infertility. Subsequently, the patient underwent testicular biopsy. Histopathological results confirmed our presumption. After a follow up of 20 months, semen analysis was still abnormal. In this case, testicular dysplasia is the root reason for the patient's infertility. Thus, despite the benign nature of bilateral testicular tunica albuginea leiomyomas, they may cause bilateral testicular hypoplasia and infertility in men. In the case of men with fertility requirements, early local mass excision is often necessary.

Acknowledgements

We acknowledge Hayward NK, PhD for critically reading the manuscript, and thank Vincent D and Hui-Qin Liu for language editing. This work was supported by The Science and Technology Foundation of Tianjin Public Health Bureau (No. 2013KZ110).

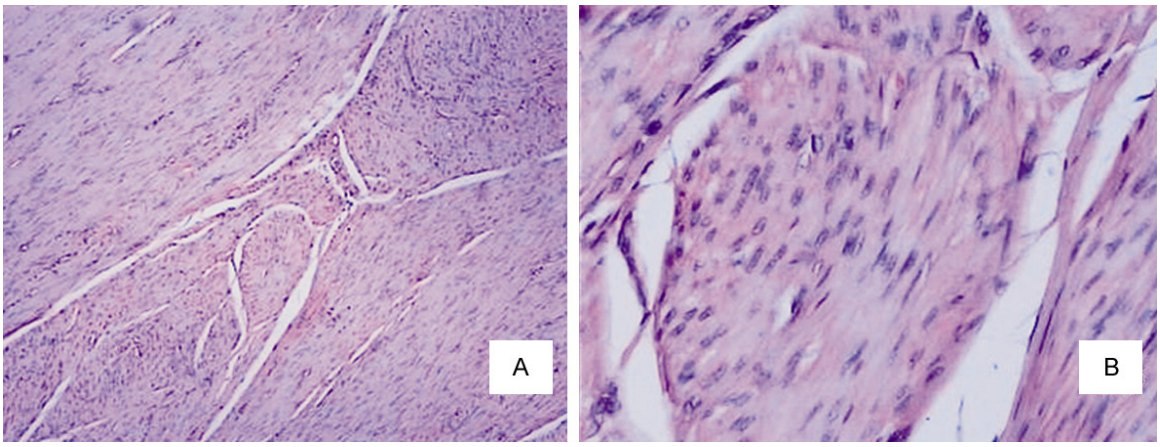

Figure 2. Histology showed interlacing uniform spindle cells with blunt-ended elongated nuclei (A, × 40; B, × 200).

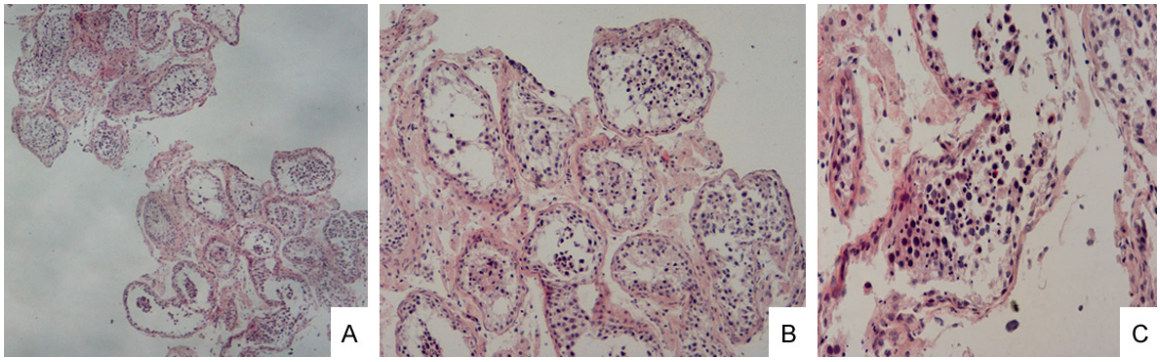

Figure 3. The number of spermatogenic cells in the seminiferous tubules were significantly decreased (A, × 40; B, × 100; C, × 200).

Disclosure of conflict of interest

None.

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