Benign fibrous pseudotumor resulting in orchiectomy despite intraoperative frozen section

Tomy Y. Perez, MD,¹ Brian J. O'Hara, MD,² Irvin H. Hirsch, MD¹ ¹Department of Urology, Thomas Jefferson University Hospitals, Philadelphia, Pennsylvania, USA ²Department of Pathology, Anatomy and Cell Biology Thomas Jefferson University Hospitals, Philadelphia, Pennsylvania, USA

PEREZ TY, O'HARA BJ, HIRSCH IH. Benign fibrous pseudotumor resulting in orchiectomy despite intraoperative frozen section. *Can J Urol* 2017;24(6):9132-9136.

Fibrous pseudotumors are rare benign lesions that originate within the paratesticular tissues. Local excision is the preferred method of treatment of these tumors over radical orchiectomy, however a definitive diagnosis must be made

Introduction

Fibrous pseudotumors are rare benign lesions that typically grow within the tunica vaginalis, but may also arise from the epididymis and tunica albuginea. They are the second most common paratesticular masses behind adenomatoid tumors, accounting for 6% of all paratesticular lesions.^{1,2} Although benign and usually asymptomatic, they often result in radical

Accepted for publication August 2017

Address correspondence to Dr. Tomy Y. Perez, Department of Urology, Thomas Jefferson University, 1025 Walnut St., Suite 1112, College Building, Philadelphia, PA 19107 USA beforehand given the similarity of these tumors to malignant entities. We present a case of fibrous pseudotumor of the tunica vaginalis and cauda epididymis. A diagnosis of fibrous pseudotumor could not be established despite the use of intraoperative frozen section, therefore necessitating radical orchiectomy.

Key Words: fibrous pseudotumor, paratesticular lesion, radical orchiectomy

orchiectomy. We present herein a case of fibrous pseudotumor and a review of the current literature.

Case report

A 48-year-old man presented with a 3 year history of an enlarging, nontender mass located in the right hemiscrotum. This was largely asymptomatic other than the occasional "twinge" sensation. The patient endorsed a history of trauma to his scrotum. Physical examination revealed a nontender, nodular, approximately 2 cm mass in the right hemiscrotum adjacent to the lower pole of the testicle. Testicles were within normal limits on exam. Scrotal ultrasound revealed a solid hypoechoic 2.1 cm x 1.1 cm x 2.3 cm mass inferior to the right testicle, as well as multiple solid nodules superior to the testis, the largest measuring 1.4 cm, and small bilateral hydroceles. Tumor markers were all within normal limits.

Right scrotal exploration was performed via an inguinal approach. The testis and epididymis were delivered with the surrounding tunica vaginalis. A tan colored nodular mass was encountered adjacent to the lower pole of the right testis. Multiple additional nodules approximately 5 mm-10 mm in size were encountered adjacent to the cauda epididymis and along the tunica vaginalis. The testis and epididymis were grossly unremarkable, Figures 1 and 2. Local excision of all lesions was initially performed and specimens sent for frozen section. Initial pathologic impression revealed low grade spindle cell tumor with a single mitosis, but no necrosis. Given the inability to exclude malignancy, radical orchiectomy was performed.

The surgical material was received fresh and initially examined by frozen section analysis. The tissue sample was embedded in OCT, flash frozen at -20 degrees Celsius, sectioned at 4 microns on a Thermo Micron HM550 and placed on glass slides. The slides were stained using a modified hematoxylin and eosin method



Figure 1. Nodules of the tunica vaginalis. **A)** Fibrous surface nodules with scattered chronic inflammation (H&E 2x).



Figure 2. Nodule in Cauda Epididymis. **A)** Fibrous nodule (H&E 1x).

and examined by a pathologist. The remaining material was fixed in 10% formalin, processed on a Thermo-Shandon tissue processor and embedded in paraffin. Tissue blocks were cut at 4 microns on a Reikert –Jung microtome, placed on glass slides and stained with hematoxylin and eosin (H&E). Immunohistochemical stains were performed using the Ventana Ultra platform.

The tunica vaginalis specimen was grossly studded by multiple white firm nodules ranging in size from a few millimeters to 0.5 cm. with areas of coalescence. The cauda epididymis specimen contained a 3.6 cm solid firm white mass. Histologically, the tumor was comprised of a low grade spindle cell proliferation with no necrosis or significant cellular pleomorphism. Only rare mitotic figures were noted. Some sections show scattered microscopic collections of chronic inflammatory cells as well as foci of hyalinization. No glandular or epithelial elements were identified. Immunohistochemical stains (calretinin, CD34, smooth muscle actin, calretinin, beta catenin) were negative within the tumor bed. An epithelial marker (AE1/AE3) showed positive staining in only surface and reactive submesothelial cells. The overall histologic and immunohistochemical features are consistent with a benign fibrous/inflammatory pseudotumor of the paratestis.

TABLE 1. Ov	erview of	fibrous	pseudotumor	cases
-------------	-----------	---------	-------------	-------

Case	Age	Predisposing factors	Symptoms	Exam	Radiologic findings	Structures involved	Frozen section	Procedure
Ugras et al ⁴	27	Left varico- celectomy	4y hx palpable nodules	Multiple, firm, tender nodules in adnexa	US: normal testes; solid, round, 5-13mm hypoechoic lesions adjacent to L testis	Tunica albuginea, tunica vaginalis, epidydimis	Ν	Radical orchiectomy
Ugras et al ⁴	37	UTI	20d hx scrotal swelling	Palpable nodules	US: homogenous multiple paratesticular nodules	Tunica albuginea	Ν	Radical orchiectomy
Parker et al ¹⁰	21	Trauma	4y hx irregular, asymptomatic mass	Firm nontender irregular mass	US: tunica vaginalis mass	Tunica vaginalis	Ν	Local excision
Tirtayasa et al ⁶	59	Inguinal hernia repair	3m rapidly growing painless enlargement	Firm, nontender mass	MRI: R inguinal hernia, hydrocele	Tunica vaginalis	Ν	Radical inguinal orchiectomy
Park et al ⁹	33	Recurrent epididymitis	Scrotal swelling and palpable mass	Firm nodular mass	US: diffuse enlargement of epididymis	Epididymis	Ν	Radical orchiectomy
Zhang et al⁵	34	None	10y hx asymptomatic scrotal mass	n/r	US: R epididymis nodule	Epididymis	Ν	Local excision
Zhang et al⁵	38	None	1y hx asymptomatic scrotal mass	n/r	US: L epididymis nodule	Epididymis	Preop biopsy- adenom	Local excision a
Zhang et al⁵	26	None	2y hx asymptomatic scrotal mass	n/r	US: Multiple L tunica vaginalis nodules	Tunica vaginalis	Frozen section fibrous pseudot	Local excision tumor
Zhang et al⁵	21	None	2y hx asymptomatic scrotal mass	n/r	US: diffuse incrassation of tunica vaginalis, testicular atropy, hydrocele w/ multiple calcificatio	Tunica vaginalis ns	N	Radical orchiectomy
Zhang et al⁵	60	None	3y hx asymptomatic scrotal mass	n/r	US: diffuse incrassation of tunica vaginalis, testicular atrophy, hydrocele	Tunica vaginalis	Ν	Radical orchiectomy
Valsero et al ⁷	34	Right inguinal hernia repair	4y hx uncomfortable right scrotal swelling	Right hydrocele, scrotal mass	US: complicated R hydrocele w/ thickened tunica vaginalis	Epididymis, tunica vaginalis	Ν	Local excision
Al-Sayyad et al ⁸	31	n/r	6m hx enlarging asymptomatic scrotal mass	Firm, nodular nontender scrotal mass	US: R epididymis mass	Epididymis, tunica vaginalis	Frozen section no maligna	Local excision incy

Case	Age	Predisposing factors	Symptoms	Exam	Radiologic findings	Structures involved	Frozen section	Procedure
Polsky et al ¹	29	None	5y hx enlarging palpable nodules w/ pain	Firm, nontender nodules	US: small hydrocele, several hypoechoic lesions adjacent to R testicle	Epididymis, tunica albuginea, tunica vaginalis	Frozen section benign fibrosis	Local incision, local excision
Tobias- Machado et al ³	60	Trauma	6y hx increasing painless swelling	Fibroelastic, nontender mass; hardened epidydimis	US: homogenous testicular mass, heterogenous epididymis	Epididymis, tunica albuginea	Frozen section fibroma versus fibrosar	Inguinal incision, radical orchiectomy coma
Seethala et al ²	57	Mumps orchitis	Firm, irregular mass	Firm, nontender mass	US: complex lesion encasing testicle, complex hydrocele	Epididymis, tunica albuginea, tunica vaginalis	Ν	Radical orchiectomy

Discussion

Fibrous pseudotumors were first recognized in 1904 by Balloch.² Two thirds of cases arise from the tunica vaginalis, 10% from the epididymis, and rarely from the tunica albuginea or spermatic cord.³ Variable nomenclature has been used previously in the literature, including fibroma, nonspecific peritesticular fibrosis, reactive periorchitis, fibromatous periorchitis, chronic proliferative periorchitis, and inflammatory pseudotumor.²

Fibrous pseudotumor is a benign inflammatory reaction. Although the precise etiology is unknown, patients often report a history of infection, trauma, or hydrocele, which suggests a reactive etiology. Hydrocele is the most frequently associated finding, with some authors reporting the association in nearly 50% of cases of fibrous pseudotumor.⁴ Our patient reported a history of trauma and was found to have hydrocele on ultrasound. Most patients present with an enlarging, painless scrotal mass that has been present for several years. They can present at any age, but the peak incidence is the third decade of life.⁵

Physical examination often reveals a single nontender mass, or multiple nodules within the scrotum. Ultrasound may reveal single or multiple hypoechoic lesions, at times associated with hydrocele, although these findings are not specific.^{6,7} Histologically, paucicellular, hyalinized fibrous tissue with some inflammatory cells is characteristic for fibrous pseudotumor.^{4,8}

Fibrous pseudotumors clinically mimic malignancy, therefore it is important to rule out other possibilities such as fibrous mesothelioma, fibroma of the tunics, inflammatory myofibroblastic tumor, neurofibroma and idiopathic fibromatosis. The most important of these would be fibrous mesothelioma. Malignant mesothelioma usually occurs in the pleura (75%) followed by the peritoneum and rarely in other sites including the scrotal region. Most malignant mesotheliomas are of the epithelioid histologic subtype while the fibrous form is the least common. A battery of immunohistochemical stains are often employed to rule out the various entities. In this case, none of the mesothelial markers were positive in the tumor bed and the negative CD34 stain did not support fibromatosis. The presence of both the fibrosis and scattered collections of chronic inflammatory cells also points to the diagnosis of fibrous/inflammatory pseudotumor of the paratestis.

Of 16 cases reviewed in the literature, nine (56%) resulted in radical orchiectomy, Table 1. In those cases where intraoperative frozen section was obtained, only local excision of the lesion was required. Our case differs in that despite obtaining a frozen section, radical orchiectomy was performed due to remaining uncertainty of the diagnosis. This finding elucidates the difficulty in making a clear diagnosis of benign fibrous pseudotumor both pre and intraoperatively. Treatment of choice remains local mass excision in order to preserve both fertility and body image of patients. Improved intraoperative diagnostic modalities are needed in order to attain this goal.

Benign fibrous pseudotumor resulting in orchiectomy despite intraoperative frozen section

References

- Polsky E, Ray C, Dubelier L. Diffuse fibrous pseudotumor of the tunica vaginalis, testis, epididymis and spermatic cord. J Urol 2004;171(4):1625-1626.
- 2. Seethala R, Tirkes A, Weinstein S et al. Diffuse fibrous pseudotumor of the testicular tunics associated with an inflamed hydrocele. *Arch Pathol Lab Med* 2003;127(6):742-744.
- 3. Tobias-Machado M, Lopes Neto AC, Simardi LH et al. Fibrous pseudotumor of tunica vaginalis and epididymis. *Urology* 2000; 56(4):670-672.
- 4. Ugras S, Yesil C. Fibrous pseudotumors of tunica albuginea, tunica vaginalis and epididymis: report of two cases. *Cancer Epidemiol* 2009;33(1):69-71.
- 5. Zhang Z, Yang J, Li M et al. Paratesticular fibrous pseudotumor: a report of five cases and literature review. *Front Med* 2014;8(4): 484-488.
- 6. Tirtayasa PM, Birowo P, Putrano AS et al. Fibrous pseudotumor of the tunica vaginalis associated with hydrocele and testicular atrophy. *Urol Case Rep* 2014;2(4):134-136.
- Valsero ME, Samaniego MP, Lagarto EG et al. Fibrous pseudotumor affecting the tunica vaginalis, epididymis and seminal duct. *Arch Esp Urol* 2013;66(9):873-877.
- Al-Sayyad A, Cagiannos I. Fibrous pseudotumor of the epididymis and tunica vaginalis. *Can J Urol* 2006;13(5):3279-3280.
- Park BK, Kim SH, Moon MH. Fibrous pseudotumor of the epididymis: sonographic and pathologic correlation. *Eur J Radiol* 2003;46(2):53-55.
- 10. Parker PM, Pugliese JM, Allen Jr RC. Benign fibrous pseudotumor of tunica vaginalis testis. *Urology* 2006;68(2):427.e17-e19.