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Testis-Sparing Surgery for Multiple Fibrous Pseudotumors of the Epididymis: A Case Report

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Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Patient: Male, 44-year-old
Final Diagnosis: Testis-sparing surgery for multiple fibrous pseudotumors of the epididymis
Symptoms: History of multiple firm, nontender left epididymal nodules. Scrotal ultrasonography showed multiple hypoechoic lesions without testicular invasion, and tumor markers were negative
Clinical Procedure: —
Specialty: Urology
Objective: Rare disease
Background: Fibrous pseudotumor of the epididymis is a rare benign reactive lesion that accounts for approximately 10% of paratesticular fibrous pseudotumors. Its etiology is unclear but is often associated with trauma or inflammation. Although it is the second most common paratesticular tumor after adenomatoid tumor, its epididymal origin is particularly uncommon. The nonspecific clinical and imaging features frequently lead to a preoperative misdiagnosis of malignancy, potentially resulting in unnecessary radical orchiectomy.
Case Report: We present the case of a 44-year-old man with a 4-year history of multiple firm, nontender left epididymal nodules. Scrotal ultrasonography showed multiple hypoechoic lesions without testicular invasion, and tumor markers were negative. Intraoperatively, the masses were well-demarcated from the testis, with an intact tunica albuginea, enabling direct testis-sparing complete resection; frozen section was deemed unnecessary. The resected specimen comprised an aggregate 8-cm mass of gray-white nodules. Histopathology revealed a fibrous pseudotumor composed of proliferating fibroblasts and myofibroblasts in a storiform pattern, accompanied by prominent lymphoplasmacytic infiltration. Immunohistochemistry showed an IgG4/IgG ratio < 40%, excluding IgG4-related disease. The differential diagnoses of adenomatoid tumor, inflammatory myofibroblastic tumor, and paratesticular sarcoma were ruled out. The patient recovered uneventfully, and at 3-, 6-, and 12-month follow-up no recurrence was observed, with normal testicular morphology and blood flow.
Conclusions: This case underscores that when multiple epididymal masses are well-demarcated from the testis, fibrous pseudotumor should be considered, enabling testis-sparing surgery and avoiding unnecessary orchiectomy. Intraoperative frozen section can be a helpful adjunct if the diagnosis is uncertain. Given the benign nature of the lesion, the long-term prognosis is excellent; nonetheless, regular ultrasonographic follow-up is recommended to monitor for potential recurrence.
Keywords: Urology • Epididymis • Fibroma • Scrotum • Orchiectomy • Case Reports
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Introduction

Fibrous pseudotumor of the epididymis is particularly rare in clinical practice. It is a paratesticular fibrous pseudotumor (PFP) arising in the epididymis and follows a benign pathological course characterized by proliferation of fibrous or collagenous tissue with hyaline degeneration, often secondary to trauma or inflammation. PFP is a rare benign tumor of unknown etiology. It originates from intrascrotal tissues such as the tunica vaginalis, epididymis, or spermatic cord [1]. The lesion typically presents as 1 or multiple nodules within the scrotum, composed of dense fibrous tissue with varying degrees of inflammatory cell infiltration [2]. Studies have reported that 76% of cases occur in the tunica vaginalis, 14% in the tunica albuginea, and approximately 10% in the epididymis [3]. Clinically, this entity must be distinguished from adenomatoid tumors and malignant tumors to avoid unnecessary orchiectomy. The present case documents a rare multiple fibrous pseudotumor of epididymal origin. The patient underwent successful testis-sparing complete resection, which clearly illustrates the importance of correctly identifying this lesion preoperatively and intraoperatively to avoid overtreatment.

Case Report

General Information

A 44-year-old man presented with a 4-year history of a progressively enlarging left scrotal mass. He reported no pain or systemic concerns. His past medical history was unremarkable, with no hypertension, diabetes, infectious diseases, trauma, drug abuse, or relevant travel history.

Examinations

Physical examination revealed multiple firm, nontender nodules in the left epididymis; both testes were normal in shape and consistency. The right testis was mildly tender, but the right epididymis and bilateral spermatic cords were unremarkable. Systemic examination was otherwise normal.

Scrotal ultrasonography demonstrated multiple hypoechoic lesions in the left epididymis, the largest measuring approximately 1.5×1.4 cm, with peripheral blood flow signals in some areas. No testicular invasion was observed. The right epididymal head appeared plump, with a suspected hypoechoic area of 0.6×0.5 cm. Laboratory results showed total prostate-specific antigen 0.63 ng/ml, alpha-fetoprotein 3.89 ng/ml, and negative tuberculosis-specific tests.

Diagnosis and Differential Diagnosis

Diagnosis: Left epididymal mass (nature to be determined).

Diagnostic basis: (1) A middle-aged male patient; (2) A left epididymal mass; (3) Physical examination revealed multiple firm, nontender nodules in the left epididymis, bilateral testes of normal shape and consistency, and no obvious thickening of the spermatic cords.

Differential diagnosis: The following conditions require differentiation. Adenomatoid tumor often presents as a solitary, small nodule; imaging findings are nonspecific, and diagnosis depends on histopathology examination [4,5]. Testicular tumors typically arise within the testicular parenchyma; ultrasonography and tumor markers can aid the initial assessment. Epididymal tuberculosis is frequently associated with a history of tuberculosis or exposure, and the vas deferens may display a beaded appearance. Inflammatory myofibroblastic tumor predominantly affects children and adolescents and is extremely rare in the reproductive system [6-8]. Scrotal skin tumors are superficially located, allowing preliminary distinction by physical examination. Additionally, malignant paratesticular tumors such as sarcoma should be kept in mind.

Treatment

After admission, preoperative examinations revealed no surgical contraindications. Following discussion with the patient and his family, left epididymal lesion resection under general anesthesia was planned. Intraoperatively, multiple masses were found. The contour of the epididymis was indistinct. The tumor bases were scattered along the tunica vaginalis wall and the testicular surface, with clear margins. The tunica albuginea remained intact, and no sign of testicular invasion was observed. Given this clearly benign appearance, the surgeons decided against submitting specimens for frozen section examination. They proceeded directly with testis-sparing complete resection of the masses. All masses were excised outside the tunica albuginea. The left testis appeared grossly normal, and the tunica albuginea was intact. The proximal left vas deferens and the spermatic cord vessels showed no tumor involvement; these structures were free of tortuosity, thickening, or stenosis (Figure 1). Intraoperatively, a cluster of gray-white nodular masses with an aggregate diameter of 8 cm was removed (Figure 2).

Postoperative pathological examination revealed that the lesion consisted of proliferating fibroblasts and myofibroblasts embedded in a fibrotic background with a storiform pattern, accompanied by abundant infiltration of lymphocytes and plasma cells (Figure 3). The immunohistochemical profile was as follows: LCA (positive in a subset of lymphocytes), CD20 (positive in a subset of lymphocytes), CD3 (positive in a subset of

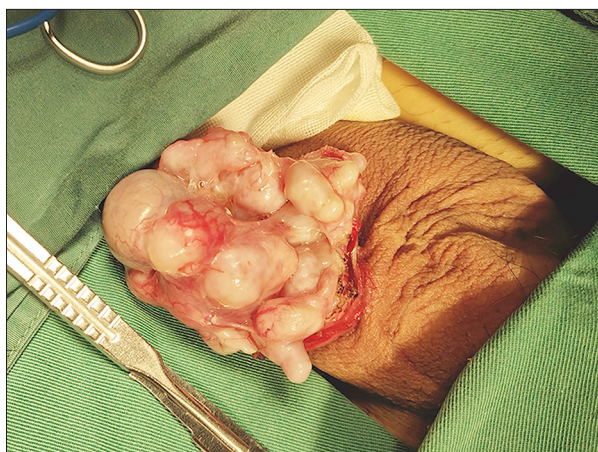


Figure 1. Intraoperative findings of the epididymis and the mass. Intraoperative photograph showing the left testis covered by an intact tunica albuginea and the residual epididymal tissue. Note the absence of tumor invasion and the clear cleavage plane between the masses and the testis.

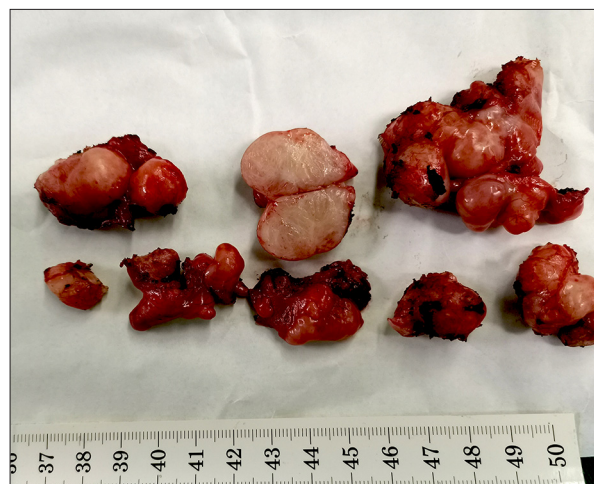


Figure 2. Excised mass specimen obtained intraoperatively. The excised tissue consists of multiple graywhite, firm, nodular masses with an aggregate diameter of 8 cm.

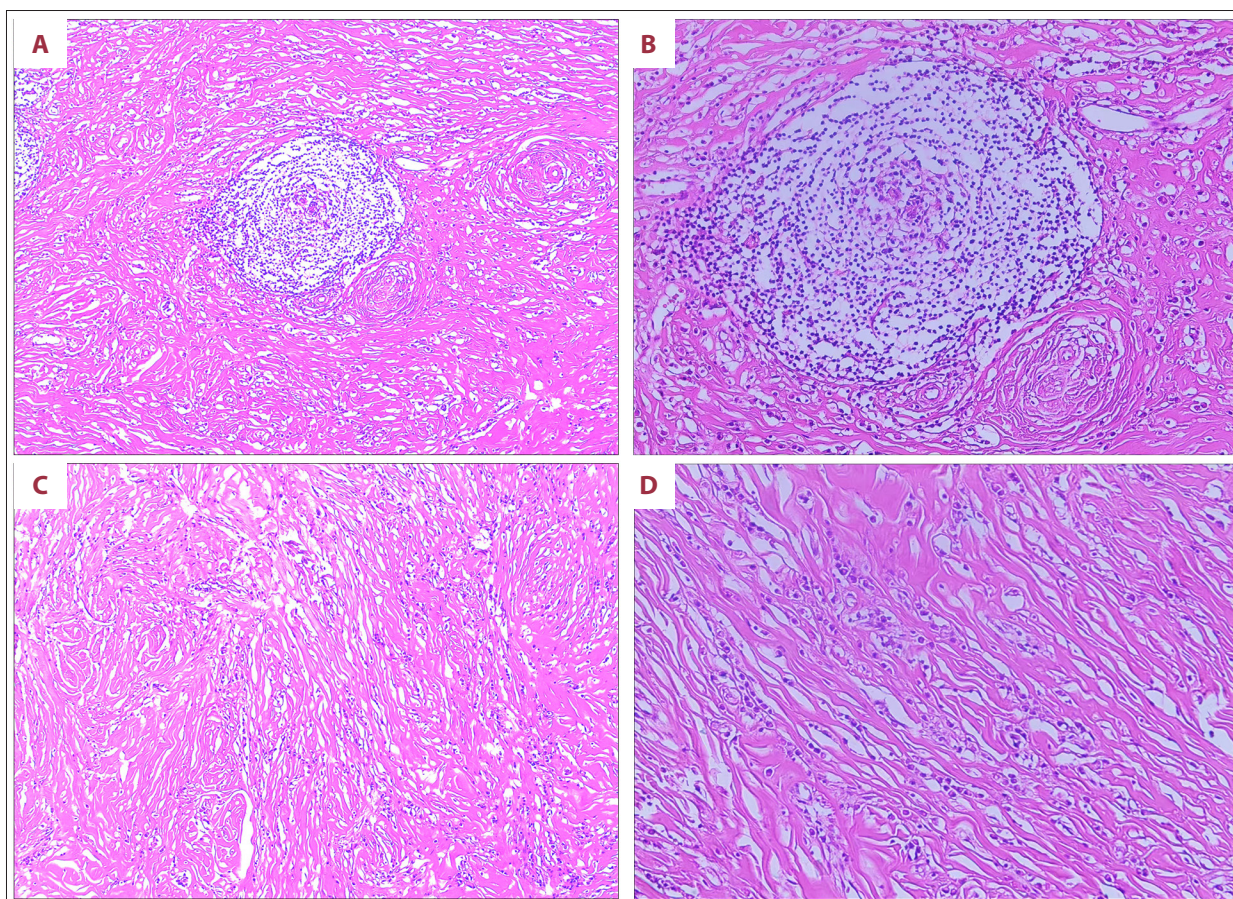


Figure 3. Histopathology images of the patient's fibrous pseudotumor of the epididymis. **A:** (HE $\times$ 100) Sheet-like proliferative fibrous tissue with focal perivascular lymphoid infiltration; **B:** (HE $\times$ 200) Demonstrating lymphoid hyperplasia; **C:** (HE $\times$ 100) Proliferating fibroblasts and myofibroblasts arranged in sheets, with marked collagen degeneration and scattered mild inflammatory cell infiltration; **D:** (HE $\times$ 200) Higher magnification of **C**.

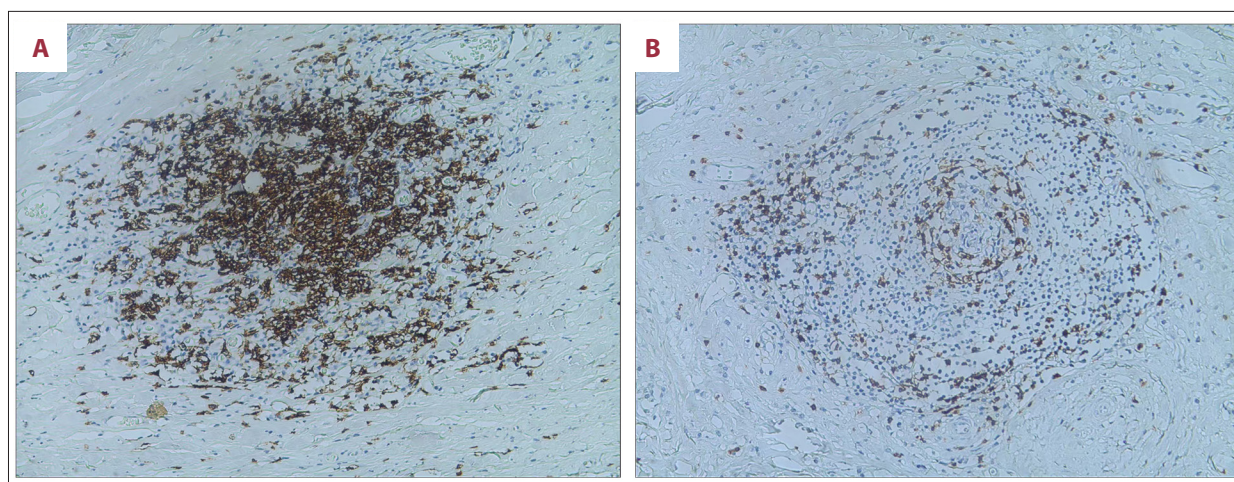


Figure 4. Immunohistochemical findings of the patient's fibrous pseudotumor of the epididymis. Brown staining indicates positive expression; CD20 and CD3 demonstrate lymphocytic infiltration. **A:** CD20 labeling B lymphocytes, $\times 200$; **B:** CD3 labeling T lymphocytes, $\times 200$.

lymphocytes), CD138 (positive in plasma cells), CD3B (positive in plasma cells), CD30 (-), SALL4 (-), CD117 (positive in mast cells), PLAP (-), CD15 (-), IgG4/IgG ratio $< 40\%$ (not meeting the common diagnostic threshold for IgG4-related disease), TP (syphilis) (-), and Ki-67 (positive in scattered cells) (**Figure 4**). The pathology diagnosis was fibrous pseudotumor of the epididymis, ruling out IgG4-related disease.

Treatment Outcome, Follow-up, and Prognosis

The wound healed uneventfully, and postoperative recovery was smooth. Follow-up examinations at 3, 6, and 12 months after surgery, including scrotal ultrasonography, revealed no evidence of recurrence. Testicular morphology and blood flow remained normal. The patient currently reports no pain or scrotal heaviness and shows no clinical signs of testicular dysfunction, consistent with a favorable prognosis.

Discussion

Paratesticular tumors are rare intrascrotal neoplasms arising from the connective tissues of the spermatic cord, epididymis, or adjacent to the testis [9]. Among them, PFP accounts for approximately 6% of paratesticular lesions [10]. It is the second most common paratesticular lesion after adenomatoid tumor, with an extremely low incidence, mainly affecting young men aged 20 to 40 years [2]. The disease typically presents as an intrascrotal mass and can originate from the tunica vaginalis, epididymis, or spermatic cord.

First described as a fibrous lesion by Astley Cooper in 1830, the entity was later designated fibrous pseudotumor by Mostofi and Price in 1973 to emphasize its reactive, benign nature [3,11].

Regarding the etiology, Mostofi observed that 30% of patients have a history of trauma or inflammation, and Sajjad et al suggested that epididymitis, local surgery, trauma, or infection associated with hydrocele may also contribute. Some studies indicate that approximately half of the cases are associated with hydrocele [12-14]; however, in many cases no clear cause can be identified. Given the histopathological similarities with IgG4-related disease (IgG4-RD) [15], Bösmüller et al hypothesized that PFP is a localized manifestation of IgG4-RD [2]. Subsequent studies have proposed that PFP falls within the IgG4-RD spectrum, characterized by storiform fibrosis, obliterative phlebitis, and increased IgG4-positive plasma cells (IgG4/IgG $> 40\%$). In the present case, the IgG4/IgG ratio was $< 40\%$ and the typical histological triad was absent, ruling out IgG4-RD [16,17].

The size of PFP nodules ranges from 0.5 cm to 8.0 cm [18]. The lesion can be diffuse or nodular, and the tunica vaginalis can exhibit diffuse thickening. Histopathology reveals proliferation of fibrous and collagenous connective tissue. Most collagen fibers arrange themselves in parallel arrays, with lymphocytes and plasma cells visible in the center. Some fibrous pseudotumors contain abundant fibroblasts and myofibroblasts. In long-standing lesions, the collagen undergoes fusion, hyaline degeneration, and inflammatory cell infiltration. The fibrous tissue assumes a storiform arrangement. Calcification or ossification can develop within the lesion in some cases. These features can make differentiation from inflammatory pseudotumor challenging, and some authors consider them different stages of the same inflammatory process [19].

In the present case, the PFP originated from the epididymis, a presentation accounting for only 10% of reported cases. Fibrous pseudotumors lack specific clinical symptoms, and their pathophysiology remains incompletely understood [20].

Patients typically present with a painless scrotal mass, sometimes accompanied by epididymal distending pain; physical examination reveals a firm, well-defined, nontender mass [21]. In our patient, the multiple nodules mimicked a malignant tumor, but intraoperative findings of clear demarcation and intact tunica albuginea allowed testis-sparing resection. This experience shows that even multinodular lesions well-demarcated from the testis should raise suspicion for a benign process, thus avoiding unnecessary orchiectomy. Diagnosis mainly relies on pathology, and the differential diagnosis includes adenomatoid tumor, inflammatory myofibroblastic tumor, epididymal tuberculosis, and paratesticular sarcoma [22-24]. Intraoperative frozen section examination can assist in determining the surgical extent when the nature of the lesion is uncertain, but it is not mandatory. When benign features are clearly recognized on gross inspection, testis-sparing resection can be performed directly, as in the present case [21,25,26]. For postoperative follow-up, regular genitourinary ultrasound examinations are recommended to detect potential recurrence early and to assess testicular morphology and function. In our patient, no recurrence was detected at the 1-year follow-up; testicular morphology and blood flow remained normal, suggesting a favorable prognosis.

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Conclusions

Fibrous pseudotumor of the epididymis is a rare, benign lesion that must be considered in the differential diagnosis of multiple paratesticular nodules to prevent unnecessary radical orchiectomy. Testis-sparing local resection, guided by intraoperative findings and frozen section examination when necessary, is the preferred management. This approach preserves testicular function and demonstrates excellent outcomes, as supported by the absence of recurrence at 1 year. The present case underscores the value of recognizing this benign entity to enable organ-preserving surgery and to avoid overtreatment.

Institution Where Work Was Done

Yichang Central People's Hospital, Yichang, Hubei, PR China.

Patient Permission/Consent Declarations

Informed consent was obtained from the patient, and the study was approved by the hospital ethics committee.

Declaration of Figures' Authenticity

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