Case Report

Uncommon Benign Intrascrotal Tumours

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Abstract

Introduction: Benign intrascrotal tumours are rare. Clinical Picture: Three patients with bilateral paratesticular leiomyomas, an adenomatoid tumour of the testis and a left paratesticular dermoid cyst coexisting with a synchronous left paratesticular epidermal cyst are presented. These tumours were discrete, hard and smooth lesions, in which the diagnosis of malignancy could not be safely excluded preoperatively even with ultrasonography. Treatment and Outcome: The first patient had orchiectomy with contralateral testicular preserving surgery, the second patient underwent orchiectomy and the third had conservative testicular surgery. Conclusion: Awareness of these benign tumours may allow for testicular preservation.

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Key words: Adenomatoid tumour, Bilateral paratesticular leiomyoma, Intrascrotal, Paratesticular dermoid cyst, Paratesticular epidermal cyst

Introduction

Leiomyomas of the urinary tract are exceedingly rare. They can, however, arise from almost any site within the genito-urinary tract, most commonly in the renal capsule and bilateral tumours are very rare.[1-3] Adenomatoid tumours are also uncommon tumours of the paratesticular tissues, and are probably of mesothelial origin.[4-8] Most adenomatoid tumours have been reported to involve the epididymis.[4-8] We present a case where this tumour was paratesticular but did not involve the epididymis. Dermoid cysts and epidermal cysts are uncommon keratinous cysts of uncertain tissue of origin.[9] We present a case where both cysts were paratesticular and coexisted in the same testis.

Case Reports

Case 1

A 64-year-old man presented with slowly enlarging bilateral scrotal swellings of 3 years' duration. There was no history of infection, trauma or other systemic symptoms. Examination revealed a large, smooth and hard right testis, approximately 10 cm in diameter. The left testis had similar characteristics, but was about 5 cm in diameter. Ultrasonography of the scrotum showed bilateral paratesticular masses of inhomogeneous echogenicity, which compressed the testes. The patient underwent right scrotal exploration. Frozen section of a wedge biopsy of the tumour revealed a leiomyoma, but a malignancy could not be excluded, so orchiectomy was performed. Exploration of the left scrotum was deferred.

Gross inspection of the specimen showed a well-defined, firm and grey-white tumour with a whorled appearance, measuring 9 x 6 x 5 cm (Fig. 1). The tumour was adherent to the tunica albuginea, compressing the testis but not invading it. Microscopic examination revealed fascicles of spindle-shaped smooth muscle cells with areas of collagenisation. The lesion was separate from the testis, outside the tunica albuginea and had no signs of malignancy. The testis was atrophic with absence of spermatogenesis. The histopathologic findings were consistent with a leiomyoma.

The left scrotal mass was observed for 2 years, during which time it slowly grew. Repeat ultrasonography showed a left scrotal mass of mixed echogenicity, with a whorled appearance. The patient agreed to surgery because of the increasing size of the tumour and underwent excision of the tumour with preservation of the testis. Gross inspection and microscopic findings were similar to that found in the right

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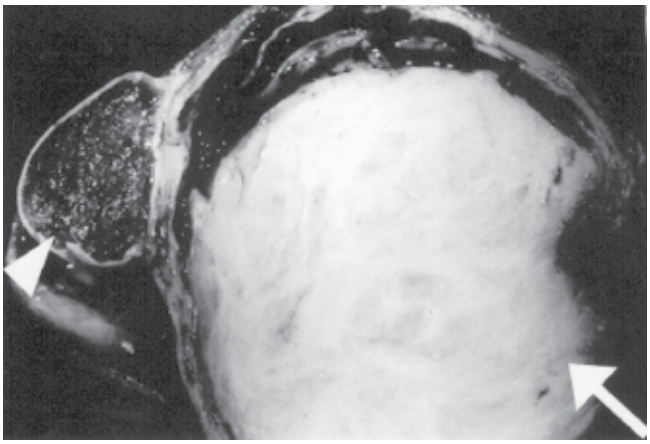

Fig. 1. Paratesticular leiomyoma. Note the well-circumscribed nature of the tumour and the whorled cut appearance. The testis (arrow head) is compressed by the leiomyoma (arrow).

testis. The diagnosis of bilateral paratesticular leiomyoma was confirmed.

Case 2

A 37-year-old man was referred for a painless left scrotal lump of several months' duration. Physical examination showed a hard and smooth left testicular mass. The patient's alpha-fetoprotein, beta human chorionic gonadotropin and lactate dehydrogenase levels were within normal limits. Computed tomography scan of the abdomen and thorax were normal.

The patient underwent a left inguinal exploration and orchiectomy. Findings were that of a pale, firm and well circumscribed mass in the lower pole of the testis measuring 3 x 3 x 2.5 cm and pushing the testis to 1 edge. The mass was separated from the testis by a densely adherent fibrous capsule. Microscopic examination showed diffuse sheets of cells with large vesicular nuclei, prominent nucleoli and abundant vacuolated cytoplasm (Fig. 2). The cells were separated by thin fibrovascular septa, with focal infiltration by lymphocytes, and were arranged in a trabecular pattern. Gland-like spaces were interspersed throughout the tumour. Immunohistochemistry with cytokeratin, epithelial membrane antigen, vimentin and S-100 were positive while carcinoembryonic antigen, factor VIII and Ber-EP4 were negative. A diagnosis of adenomatoid tumour was made.

Case 3

A 25-year-old man presented with an incidental finding of a left scrotal mass. The mass was hard and localised to the superior aspect of the left testis. Tumour markers were negative. Ultrasonography showed 2 solid nodules adjacent to the left testis measuring 4 cm and 2 cm in diameter, of similar echogenicity to the normal testicular tissue, but with areas of calcification (Fig. 3).

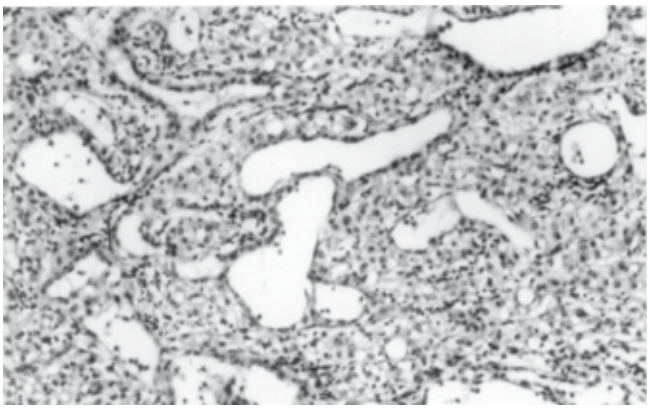

Fig. 2. Photomicrograph of the adenomatoid tumour. Neoplastic cells surround gland-like spaces. (Haematoxylin and eosin stain, x200)

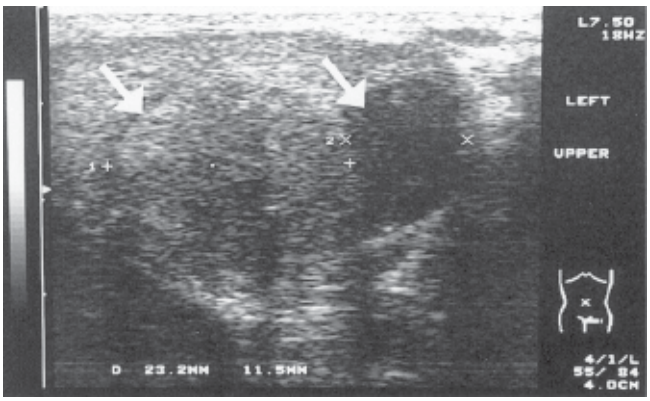

Fig. 3. Ultrasound of the left scrotum, upper pole (transverse). Image shows 2 solid nodules of different echogenicity adjacent to the testis (arrows).

An inguinal exploration, intraoperative frozen section and total excision of the left testicular lumps were performed with preservation of the left testis. Macroscopic examination revealed 2 separate, well-encapsulated paratesticular cysts adherent to the tunica albuginea of the superior aspect of the left testis. The cysts exuded cheesy material when sectioned. Histological examination showed that 1 cyst wall was lined by stratified squamous epithelium with associated sebaceous glands. The cyst contained acellular keratinous material. The diagnosis was that of a benign dermoid cyst. Microscopic examination of the other cyst revealed that the cyst wall was lined by stratified squamous epithelium with the cyst containing keratinous debris. Hence, a diagnosis of an epidermal cyst was made.

The patient was followed up clinically and with ultrasound studies. He presented 4 years later again with symptoms of pain in the left scrotum. Physical examination again revealed a lump in the middle aspect of the left testis. Tumour markers done again were within normal limits and the ultrasound study showed an irregular echogenic lesion with calcification, measuring 2 cm in diameter, in the left testis. A second inguinal exploration, frozen section and

excision of the cyst was performed. The gross and microscopic appearances were similar to the nodules excised previously. Histology confirmed a recurrence of the epidermal cyst.

Discussion

Most solid intratesticular masses are malignant. Hence, a testicular mass, especially if solid on ultrasonography, must be considered to be possibly malignant. Benign lesions, however, also do exist and recognition of this entity may allow for testicular preservation. From 1996 to 1999, 19 inguinal explorations for intrascrotal masses were performed in our institution. Histological examination, however, revealed 10 malignant and 9 benign lesions. Neoplasms of paratesticular structures, although uncommon, comprise a heterogeneous group of benign and malignant lesions. In the older male, they constitute about 15% of intrascrotal lesions.[8] About 20% of paratesticular tumours are malignant.[7] These tumours can be difficult to distinguish from intratesticular lesions on physical examination alone or even with ultrasonography. Some common intrascrotal benign pathologies are highlighted in Table 1.

Leiomyomas are benign smooth muscle tumours which although rare in the scrotum, have been found in many locations, including the epididymis (where it is the second most common neoplasm comprising 6% of all epididymal tumours), spermatic cord, tunica dartos, tunica albuginea and within the testicle.[1,2,10] Leiomyoma of the testicular tunics are extremely rare and subcapsular intratesticular leiomyomas are rarer still.[1,11] The most common time of presentation for scrotal leiomyomas is in the fifth decade of life.[2] These tumours are typically slow-growing. Bilateral leiomyomas have been reported, but are very rare.[2,3]

Our case of scrotal leiomyomas is unusual in that the right-sided tumour was so large as to cause atrophy of the testis and was tightly adherent to the tunica albuginea, thus making distinction of its origin difficult. On ultrasonography, the tumours displayed inhomogeneous echogenicity, the differential diagnosis of which, includes chronic hydrocoeles and other tumours such as adenomatoid tumours, lymphoma, germ cell tumours and fibroma.

Table 1. Possible Benign Intrascrotal Paratesticular Lesions

Location	Histological type	Significance
Paratesticular tissues, epididymis	Adenomatoid tumour	Most common paratesticular tumour (30% of paratesticular tumours) Usually confined to epididymis Presenting age: usually 20 to 30 years Usually asymptomatic Benign Presence of vacuoles within epithelial cells
Paratesticular tissues, epididymis	Leiomyoma of epididymis	Second most common paratesticular tumour May be painful and with hydrocoele associated Slow growing Usually fifth decade of life
Paratesticular tissues, epididymis	Cystadenoma of epididymis	Bilateral in 30% Associated with von Hippel-Lindau disease Young adults Asymptomatic or minimal local symptoms
Paratesticular tissues	Spermatocoele	Reasonably common Represents a cystic accumulation of semen in the epididymis May be asymptomatic
Paratesticular tissues	Chronic epididymo-orchitis	May have chronic scrotal pain, Induration of epididymis May have history of acute epididymo-orchitis
Intratesticular	Epidermoid cyst	1% of testis tumours Presenting age: between teens and thirties, usually in twenties Benign Well circumscribed on ultrasound
Intratesticular	Adenomatoid tumour	Uncommon Usually small, benign Fibrous stroma with disoriented spaces of epithelial cells
Intratesticular	Specific inflammations (infections)	Tuberculosis, mumps, and syphilis are considered. Correlation between clinical, radiological and microbiological features is crucial

Exploration was indicated as there was a need to exclude malignancy. Excision of the left testicular lesion was delayed as it had similar physical and ultrasonic features as the right tumour, and the patient was not keen on surgery initially. Preservation of the left testis had preserved the patient's potency.

Adenomatoid tumours are rare, benign tumours that primarily occur in the paratesticular tissues of males and the uterus and fallopian tubes of females.[4,5] They are reported to be the most common tumour of the testicular adnexa; in 1 report, they accounted for approximately 30% of all paratesticular tumours.[4,6-8] The majority are found in the epididymis, and rarely does the tumour originate in the testicular tunica, spermatic cord, ejaculatory ducts, prostate or suprarenal areas.[4,5,7] The cytological origin of adenomatoid tumours has been a source of controversy, but most believe it to be of mesothelial origin.[4-8] They usually present in the third to fifth decades of life as an incidental hard, painless mass in the scrotum.

Our case of adenomatoid tumour conformed to its classical clinical presentation. Pre-treatment biopsies are regarded to have no role in the management of testicular tumours; exploration and histological examination can be regarded as mandatory.[4] In retrospect, a frozen section may have allowed for testicular preservation. Some have suggested that these tumours can be treated by local excision, as there has been no report of recurrences.[4]

Keratinous cysts of the testis are rare, are usually intratesticular and account for only 1% of testis tumours.[9] The epidermoid variant of keratinous cyst is distinguished from the dermoid cysts by the lack of adnexal skin structures.[9] Dermoid cysts occur in many different tissues in the body, but are extremely rare in the testis. A testicular dermoid cyst may actually be a bilayer expression of a teratoma.[12] The cysts usually present during the third decade of life. These lesions are well-encapsulated and are unilocular and do not metastasize.[12] Our case of dermoid cyst was unique because the cyst was paratesticular and coexisted with a separate paratesticular epidermal cyst within a single testis.

Epidermal cysts are usually asymptomatic and detected during self-examination or routine physical examination. The majority of the patients are in the second to fourth decades of life.[13] An epidermal cyst usually appears as a well-circumscribed, smooth, firm and non-tender lump. They are usually reported to be intratesticular where the term epidermoid cysts have been used.[12-14] Ultrasonography may show a circumscribed hypoechoic lesion with hyperechoic margins.[13,15] Its histogenesis is not known, and most consider the origin to be a strictly monomorphic ectodermal development of a teratoma along the line of epidermal differentiation, or less likely the squamous metaplasia of the seminiferous epithelium or rete testis.[13] Treatment is controversial with previous literature advocating inguinal orchiectomy, but there have been increasing number of reports in recent years favouring organ-sparing surgery.[13] A clinical approach to scrotal masses is shown in Figure 4.

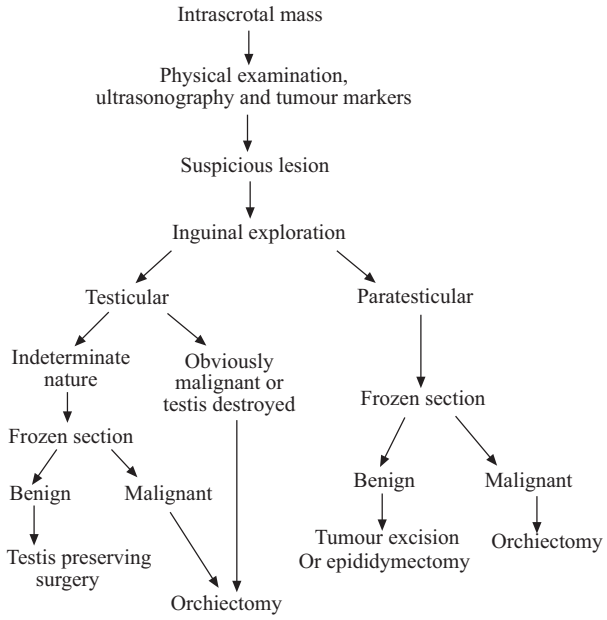

Fig. 4. Clinical approach to suspicious intrascrotal lesions.

Conclusion

Ultrasonography is not always helpful in distinguishing a paratesticular lesion from an intratesticular lesion. A paratesticular lesion should alert us to the possibility of the mass being benign. Nevertheless, inguinal exploration of suspicious scrotal masses is mandatory. We recommend routine intraoperative frozen section biopsy as it may allow for testicular preservation.

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