Intrascrotal adenomatoid tumour: A report of seven cases and review of the literature

Tomasz Golabek¹, Jakub Bukowczan², Tomasz Wiatr¹, Mikolaj Przydacz², Lukasz Belch¹, Piotr Chlosta³

¹Department of Urology, Collegium Medicum of the Jagiellonian University, Krakow, Poland
²Department of Endocrinology and Diabetes Mellitus, Diabetes Resource Centre, North Tyneside General Hospital, North Shields, United Kingdom

Abstract

Adenomatoid tumours (ATs) are rare intra-scrotal neoplasms. Although a diagnosis of this type may be suspected when found in the epididymis, an intra-testicular location can cause a diagnostic and therapeutic dilemma. Thus, we report our experience with intra-scrotal adenomatoid tumours - 5 epididymal, 1 intra-testicular, and 1 involving the epididymis and testis, as well as a literature review to better understand this benign but clinically significant lesion.

Introduction

Adenomatoid tumours (AT) may be found in both male and female genital tracts, but are generally considered uncommon findings. In men, they usually arise in the epididymis but may also involve the testicular tunica and spermatic cord². In women, these benign neoplasms have been located in the uterus, fallopian tubes, and rarely in the ovary². The origin of adenomatoid tumours has been a controversy for many years, however, comparative electron microscopic and immunohistological studies have demonstrated a mesothelial origin³. In most cases of adenomatoid tumour involving the epididymis, the diagnosis may be strongly suspected as this type of neoplasm represents the most common lesion in that particular location³. Additionally, an asymptomatic course and an incidental finding of a small slow growing and painless mass adjacent to the testis, are both in favour of such diagnosis. Nevertheless, in some cases, especially when it originates from within the tunica albuginea with secondary involvement of the adjacent testicular parenchyma, AT may be indistinguishable from a malignancy and may give rise to a clinical quandary.

In this article, we present a retrospective analysis of seven cases of adenomatoid intrascrotal tumours, and we also include a brief literature review.

Material and methods

All male patients who underwent surgery for a scrotal mass between August 1984 and July 2014 were identified using the hospital patient registration...
Intrascrotal adenomatoid tumour, p. 42 - 46

database and histology registry database. Patients with histologically - proven adenomatoid intrascrotal tumours were included for analysis. A retrospective review of the charts confirmed the diagnosis.

The following data were collected and selected for analysis: age, presenting complaints, location of the lesion, initial and final diagnosis, type of procedure, and complications.

**Results**

The clinical data of all men with benign adenomatoid tumours are shown in Table 1.

An incidence of 1.6 per 100,000 hospital admissions was found. There were 7 intrascrotal ATs diagnosed between 1984 and 2014. Of these, one was located within the testis, five within the epididymis and one within the testis and epididymis.

Adenomatoid tumours comprised 26.3% of all epididymal neoplasms and they represented 0.85% of all testicular tumours. All patients were of Caucasian origin. Their age ranged from 34 to 69 years (mean 45 years). Two patients were symptomatic and complained of intermittent scrotal pain which resolved following the surgery. Four tumours were found incidentally by the patients themselves after self - examination and one was discovered at hydrocelectomy. The time from the discovery of a scrotal mass by a patient to seeking a medical attention ranged from 4 months to 2 years and it was shorter in two patients who had a history of previous blunt scrotal injury. There was no familial incidence of intrascrotal tumours recorded, and all patients, except one who had a 2 - year history of hypertension, had no other medical conditions.

On physical examination, a non - tender mass was noted in all cases. It was confined to the tail of the epididymis in four patients, to the head of the epididymis and upper pole of testis in one patient, and to the lower pole of the testis in one another. A hydrocele was diagnosed in one case.

Three patients had local excision of the tumour, one had a partial epididymectomy, one had a total epididymectomy, and one had a simple orchidectomy. The youngest patient who had a short history of a testicular mass underwent radical orchidectomy. No recurrence was found during the follow - up ranging from one to three years.

**Discussion**

Adenomatoid tumour of the male genital tract has been recognised since 1945. It comprises 32% of all paratesticular tissue neoplasms and it is the most common benign epithelial tumour of the epididymis representing up to 77% of all cases. ATs may be found in the tunica albuginea of the testis in approximately 14% of cases and, rarely, in the spermatic cord, prostate, ejaculatory ducts and scrotal capes.

The reported incidence 1 - 2 per 100,000 admissions, which is similar to our findings, seems to be an underestimation, considering its frequent asymptomatic course. Moreover, the misdiagnosis of a spermatocele could clearly affect its true prevalence.

Although the tumour may be found anywhere worldwide, there are significant geographic variations in the reported incidence rates with Caucasian men being the most commonly affected group (9:1). Adenomatoid tumours can occur across all ages, however, they remain most commonly diagnosed in the third and fourth decades of life. There is no predilection to a particular side, confounding reports regarding the predominant location within the epididymis exist, however. In our series, the tail of the epididymis was involved in three cases, while the head of the epididymis was involved in two patients.

An intra - testicular location of AT occurs only exceptionally with no more than eleven cases involving the testicular parenchyma having been reported so far. In our series, only one out of seven men had a tumour located within the testicular parenchyma.

Clinically, adenomatoid tumour presents as a slow growing, small, solid, well - demarcated mass, without a capsule nodule and which does not transilluminate. In older men it is usually asymptomatic and found on routine examination or at time of surgery. On the other hand, in younger patients, a more rapidly progressing mass which is often associated with pain seems to be a common manifestation. A similar pattern of presentation was found in our series. The observed differences could likely be explained by more frequent self - examination and a greater awareness among rather young males but also by possibly different natural history.

Approximately 20% of ATs are associated with the hydrocele. In these cases, this benign neoplasm is found...
Intrascrotal adenomatoid tumour, p. 42 - 46

Incidentally following hydrocelectomy. Trauma of the scrotum preceding discovery of the tumour has been reported in few patients, giving rise to a controversy associated with its histogenesis. Still, comparative electron microscopic and immunohistological studies have demonstrated similarities with the mesothelium, therefore the association with trauma seems rather anecdotal. Nonetheless, two of our patients had blunt scrotal injury 3 to 5 months before the discovery of a lesion. Five percent of men present with symptoms suggesting acute epididymitis.

Scrotal ultrasonography is a recommended preoperative imaging modality as it determines the location and differentiates cystic from solid lesions. However, in cases of intratesticular ATs, its usefulness is limited since they may appear as hypo-, iso-, or hyperechoic lesions. Likewise, the Nuclear Magnetic Resonance imaging may not provide the definitive diagnosis.

|| Case | Age (yrs) | Presenting symptoms | Clinical findings | Other studies | Procedure | Histology |
|---|---|---|---|---|---|---|
| 1 | 34 | Incidental finding, 1 year | Small, hard, non-tender mass of the head of the right epididymis | Frozen sections: no evidence of malignancy | Partial epididymectomy | Adenomatoid tumour of the epididymis |
| 2 | 37 | Incidental finding, 2 years | Small, hard, non-tender mass in the inferior pole of the left testis | BHCG, AFP, LDH: normal; Frozen sections: no evidence of malignancy | Local excision | Adenomatoid tumour of the testis involving tunica albuginea |
| 3 | 48 | Intermittently painful, small scrotal mass, 5 months; prior scrotal injury | Small, hard, non-tender mass of the left epididymal tail | No tests performed | Local excision | Adenomatoid of the epididymis |
| 4 | 69 | Intermittently painful, large scrotal mass, 1 year | Large 9x6x5 cm, non-tender, right hydrocele; Hydrocele and 6 cm mass attached to the right epididymis found at surgery | No tests performed | Scrotal orchidectomy | Right hydrocele and adenomatoid tumour of the right epididymis |
| 5 | 40 | Incidental finding, 6 months, prior scrotal injury | Small, hard, non-tender mass of the right epididymal tail | US: calcified haematoma in the right epididymis | Epididymectomy | Adenomatoid of the epididymis |
| 6 | 49 | Incidental finding, asymptomatic, 2 years, increase in size over 1 year | Small, hard, non-tender mass of the right epididymal tail | US: 3x1 cm hypo-echoic tumour | Local excision | Adenomatoid of the epididymis |

Key: yrs = years; BHCG = Human Chorionic Gonadotropin; AFP = Alpha Fetoprotein; LDH = Lactate dehydrogenase; US = scrotal ultrasound

TABLE 1

Physical, clinical and radiological characteristics of the patients with benign adenomatoid tumours

incidentally following hydrocelectomy. Trauma of the scrotum preceding discovery of the tumour has been reported in few patients, giving rise to a controversy associated with its histogenesis. Still, comparative electron microscopic and immunohistological studies have demonstrated similarities with the mesothelium, therefore the association with trauma seems rather anecdotal. Nonetheless, two of our patients had blunt scrotal injury 3 to 5 months before the discovery of a lesion. Five percent of men present with symptoms suggesting acute epididymitis.

Scrotal ultrasonography is a recommended preoperative imaging modality as it determines the location and differentiates cystic from solid lesions. However, in cases of intratesticular ATs, its usefulness is limited since they may appear as hypo-, iso-, or hyperechoic lesions. Likewise, the Nuclear Magnetic Resonance imaging may not provide the definitive diagnosis.

Adenomatoid tumours have no malignant potential, and no cases of metastasis or recurrence after excision have been reported. Treatment, therefore, is by surgical excision. If a benign lesion of the testis is suspected, a fine needle aspiration cytology or an intra-operative biopsy should be performed prior to dividing the spermatic cord. This approach allows to avoid unnecessary orchidectomy. Cytological features of AT include smears containing sheets of epithelial cells and clusters of monomorphic cells with round or oval nuclei and inconspicuous nucleoli. Cytoplasm is clear and vacuolated in the Papanicolaou stain and stains pink with the Giemsa stain. Naked nuclei and
fragments of stroma along with few lymphocytes can also be seen. 

Postoperative histological examination provides the final diagnosis. Microscopically, adenomatoid tumours are characterized by three basic patterns: tubules, cords, and small nests lined by, or formed of, cells that are cuboidal with moderate to abundant basophilic, eosinophilic, or vacuolated cytoplasm. The stroma contains fibroblasts, blood vessels and smooth muscle and is usually fibrous and occasionally hyalinized.  

In view of the range of microscopic appearances of adenomatoid tumours, diagnostic problems may arise in differentiation between the Sertoli or Leydig cell tumours, liposarcoma, mesothelioma, yolk sac tumours, and metastatic adenocarcinoma. In the confounding cases, immunohistochemical confirmation with mesothelial-related markers (calretinin, CKS/6 and WT - 1) is used to distinguish adenomatoid tumours from the non-mesothelial lesions, even in the presence of infarction.

In conclusion, the adenomatoid tumours are uncommon lesions that may cause a clinical dilemma, especially when the location is intra-testicular. Preoperative imaging studies remain rather unhelpful in differentiating between the benign and malignant lesions. Under these circumstances, a radical surgical approach is warranted. However, if a benign neoplasm is suspected, no surgery, or perhaps a modified procedure allowing for the testicular preservation, is appropriate.

Περίληψη

Οι αδενωματώδεις όγκοι του οσχέου είναι σπάνια νεοπλάσματα. Αν και μια υπόνοια διάγνωσης αυτού του τύπου μπορεί να τεθεί όταν ένα νεόπλασμα με παρόμοια χαρακτηριστικά βρεθεί στην επιδιδύμιδα, όταν εντοπίζεται στον όρχι μπορεί να προκαλέσει διαγνωστικό και θεραπευτικό δίλημμα. Στο παρόν άρθρο αναφέρουμε την εμπειρία μας με πέντε επιδιδύμιδες, ένα τον όρχη και έναν που εντοπίζονταν ταυτόχρονα και στον όρχη και στην επιδιδύμιδα. Επιπλέον παρατίθεται μια επισκόπηση της βιβλιογραφίας για την καλύτερη κατανόηση αυτής της καλοήθους αλλά κλινικά σημαντικής οντότητας.

Λέξεις ευρετηριασμού

αδενωματώδης όγκος, καλοήθης όγκος της επιδιδύμιδας, ενδο-οσχεικό νεόπλασμα
References