Intra-Scrotal Adenomatoid Tumour Referred as an Extra Testis: A Case Report and Review of the Literature

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Abstract

“Background:”
Intra-scrotal adenomatoid tumours are rare tumours which may present at any age and are usually treated by local excision. Ultrasound scan may help in the diagnosis but its capability to distinguish this lesion is low in view of the non specific ultrasonic features of the lesion.

“Aims:”
To report a case of intrasrotal adenomatoid tumour which was referred to a urologist as atrophic third testis after ultrasound scan examination
To review the literature on adenomatoid tumours

“Case Report:”
A 48-years-old man was referred because he had noticed a painless lump in the left side of his scrotum. He had ultrasound scan of the testes and scrotal contents which was reported as showing: normal left testis and normal right testis and in addition a small solid lump in the left hemi-scrotum which the radiologist felt was most likely an atrophic extra testis (third testis).

Clinical examination revealed normal left and right testes as well as a 2 cm globular, smooth/homogenously palpable lump of firm/hard consistency abutting on the epididymis. A provisional diagnosis of adenomatoid tumour was made. The lesion was excised and its histological features were found to be consistent with adenomatoid tumour.

He presented at follow-up clinic with a residual lump in the left hemi-scrotum which after ultrasound scan was reported by the radiologist to be consistent with persistence of adenomatoid tumour. However, clinically this lump was an organised haematoma which resolved and the patient remained well without a recurrence of the lump nine months post-operatively.

“Conclusions:”
Adenomatoid tumours are rare tumours with non-specific ultra-sonographic features in view of which the unaccustomed radiologist may misdiagnose it. Haematoma is certainly one of the differential diagnoses of adenomatoid tumour. Local excision should be the recommended form of treatment which has the advantage of confirming the diagnosis. There has not been any report of recurrence after local excision of the lesion.

Introduction

Adenomatoid tumours are rare which on ultra sound scan may present as hyper-echoic, iso-echoic or hypo-echoic lesions. These features are non specific and hence a radiologist who has not encountered a case of intra-scrotal adenomatoid tumour before may not be able to confidently diagnose adenomatoid tumour based on ultra-sound scan. Supernumerary testes are rare with about 100 reported cases. We report a case of intra-scrotal adenomatoid tumour which on ultra-sound scan was mistakenly diagnosed as a possible atrophic extra testis. Literature on adenomatoid tumour has been reviewed.

Case Report

A 48-years-old man was referred because he had noticed a lump in the left side of his scrotum. He did not have any pain associated with the lump. He had ultrasound scan of the testes and scrotal contents which was reported as showing: normal left testis and normal right testis and in addition a small solid lump in the left hemi-scrotum which the radiologist was not sure of but felt it was most likely an atrophic extra testis (see illustrations 1 and 2).

On examination both his testis felt normal on palpation. In addition there was a small globular mass which felt smooth and firm, possibly attached to the head of the epididymis / vas deferens. The provisional clinical diagnosis made by the urologist was a benign adenomatoid tumour with a possible differential diagnosis of a fibroma. It was felt that the best way to confirm the diagnosis would be to have the lesion excised for histological examination.

The lump was excised under general anaesthesia and histological examination of the specimen was consistent with adenomatoid tumour (see illustrations 3 [histology of lesion] and 4[immunohistochemistry]). At the first post-operative follow-up clinic the patient...
stated that there was still a residual lump at the site of the operation. The examining doctor felt a palpable lump/nodularity and was unsure as to what the lump was. A repeat scrotal ultrasound scan was requested. The repeat ultrasound scan was reported as showing a lump which was the same size as the adenomatoid tumour and this was reported as most likely a persistent adenomatoid tumour (see illustration 5 showing a haematoma which mimic the original lesion illustrating that there are no clear cut ultrasound features which distinguish haematomata from adenomatoid tumour).

At a second follow-up 7 months after excision of the lump the patient reported that the residual lump had gradually reduced in size and completely disappeared. Examination of the scrotum and scrotal contents revealed: normal right testis and epididymis; normal left testis and epididymis; the previously palpable nodularity / lump felt in the left hemi-scrotum was no longer palpable. It was therefore concluded that the patient developed haematoma post-operatively and what the doctor palpated at the first post-operative review was an organised haematoma which had eventually resolved. It was also felt that what the radiologist thought was a persistent adenomatoid tumour was in fact organised haematoma which had resolved by the time of the second post-operative follow-up. Another scrotal ultrasound scan was requested which confirmed complete resolution of the haematoma (see illustration 6).

**Discussion**

Para-testicular tumours account for 7% to 10% of intra-scrotal masses. Seventy-five to 90% of these tumours originate from the spermatic cord; most of them (70%) are benign. Neoplasms of the epididymis and testicular tunicae are next in frequency (the latter are rare). Tumours of the epididymis may either be primary or rarely secondary.

Beccia and associates [1] reported that out of 341 epididymal tumours in their study 75% were benign. They also reported the following types of tumours among the 341 epididymal tumours:

- Adenomatoid tumours 73%.
- Leiomyomas 11%.
- Papillary cystadenomas 9%.

The remaining tumours (7%) were benign tumours of a variety of histological types (angioma, dermoid cysts, fibroma, hamartoma, teratoma, and cholesteatoma). Twenty five percent of all epididymal tumours are malignant. Of these 24% are primary epididymal carcinomas, 27% metastatic tumours and 44% are sarcomas [2].

Adenomatoid tumours are benign benign neoplasms which account for 30% of all para-testicular neoplasms. In males, adenomatoid tumours are more common in the epididymis, but they may also be found in the tunica albuginea of the testis, tunica vaginalis of the testis or on the spermatic cord. Other reported locations of the adenomatoid tumour include: the vas deferens, prostate and adrenal glands [2]. In the females, adenomatoid tumours have been reported to occur in the uterus, fallopian tube and, rarely in the ovary [3]. Adenomatoid tumours are said to be more common in the Caucasians [2]. Cases of adenomatoid tumours have also been reported in other locations including: pleura; adrenal gland; liver [4, 5]. Rare cases have been reported where the tumour had involved the testicular parenchyma, ejaculatory ducts, spermatic cord, prostate and adrenal gland.

Histogenesis of adenomatoid tumour has been explained on a variety of theories which include: Mesothelial, Mullerian, mesonephric, and endothelial origin [2]. The postulate of a mesothelial origin is the most widely accepted, and this has been supported by electron microscopy [6] showing cells very similar to those in normal mesothelium and mesothelioma and immunohistochemical studies high hyaluronic acid levels, as in mesotheliomas [7]. Adenomatoid tumours are usually diagnosed in adulthood and these have most commonly found in patients in their 3rd to 5th decades of life, however, they have also been reported in patients aged 18 to 80 years of age [2, 8]. Adenomatoid tumour usually presents as a painless mass which the patient finds incidentally. Rarely the lump may be associated with pain. Usually adenomatoid tumour presents as a small, slow-growing lesion which varies in size, ranging from 1 cm to 5 cm; predominantly located on the left side. Most adenomatoid tumours are found in the lower or upper poles of the epididymis or in their vicinity [9]. A slightly higher incidence in the lower pole has been reported [9]. An adenomatoid tumour may occasionally be associated with a hydrocele in 15% to 20% of time [9]. An ultrasound scan usually reveals the solid nature of the lesion which may be hyper-echoic, iso-echoic, hypo-echoic or it may adopt any morphology [8, 10]. It does not show significant vascularity with color Doppler studies [8]. Adenomatoid tumours present as a relatively small, well demarcated, and non-encapsulated nodule with a mean diameter of 2cm, and a reported largest diameter of 12 cm [11].

The cut surface of an adenomatoid tumour is firm and solid with variable cystic spaces. Microscopically, the tumour is comprised of two major elements,
epithelial-like cells and fibrous stroma. The epithelial-like cells are arranged in a network of tubules, cords, channels, and microcystic spaces. The fibrous stroma may be hyalinated and may contain smooth muscle. Mitotic figures are mostly absent [12]. Differential diagnoses of adenomatoid tumour include: metastatic carcinoma [7], histiocytoid hemangioma [13], and carcinoma of rete testis. Special stains with PAS with diastase and mucicarmine are negative. Alcaine blue is positive but is hyaluronidase sensitive. The cells are cytokeratin 5/6, EMA and AE1-A E3 positive, indicating the mesothelial nature of the lesion [14].

Differential diagnoses of adenomatoid tumour include: metastatic carcinoma [7], histocytoid hemangioma [13], and carcinoma of rete testis. Special stains with PAS with diastase and mucicarmine are negative. Alcaine blue is positive but is hyaluronidase sensitive. The cells are cytokeratin 5/6, EMA and AE1-A E3 positive, indicating the mesothelial nature of the lesion [14]. The cells of adenomatoid tumour are not frankly malignant on histological examination and they show hyaluronidase resistant positivity in the cytoplasm. The cells usually are CEA positive and hyaluronidase resistant in carcinoma of rete testis [14].

Immunohistochemical studies of adenomatoid tumours involving various organs have been recently shown to reveal positive staining for calretinin [15; 16; 17; 18; 19]. Our patient's adenomatoid tumour was positively stained for calretin in as shown in illustration 4.

Considering the fact that some adenomatoid tumours are hyper-echoic and others are either iso-echoic or hypo-echoic on ultrasound scan diagnosis of adenomatoid tumour may be difficult for a radiologist who has never encountered a case before. In this case the radiologist was of the opinion that the lesion may be an atrophic extra testis. The Urologist had seen five previous cases of paratesticular adenomatoid tumour before and was fairly certain of the clinical diagnosis based upon the findings of an approximately 2 cm globular /rounded, homogenously firm/hard, smooth lump abutting on the epididymis/vas, however, it is difficult to re-assure a patient of a benign pathology without histological examination of the specimen to confirm the diagnosis. Literature review indicates that adenomatoid tumour may look like a haematoma. It was therefore not surprising that when the patient developed an organised intra-scrotal haematoma post-operatively the radiological appearance of the haematoma was adjudged by the radiologist to be consistent with a persistent adenomatoid tumour. Adenomatoid tumours are benign tumours with no reported cases of recurrence after excision [9].

Conclusions

Adenomatoid tumours are benign tumours with no reported cases of recurrence or metastasis after excision. Local excision of the lesion should be the recommended form of treatment which has the advantage of confirming the diagnosis in view of the fact that ultrasound findings are non-specific and may mimic other lesions.

References

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Illustrations

Illustration 1

Illustration 1: Ultrasound scan showing a rounded lesion in the left-hemiscrotum which after excision was shown to be Adenomatoid tumour

Illustration 2

Illustration 2: Another view of ultrasound scan showing a rounded lesion in the left hemi-scrotum; this was later shown on histology to be adenomatoid tumour
Illustration 3

Illustration 3: (x20) Adenomatoid tumour showing closely packed small tubules and cords of cells (Histological picture of the lesion)

Illustration 4

Illustration 4: (x20) The Adenomatoid tumour showing positive immunostaining with calretinin
Illustration 5

Illustration 5: Ultrasound Scan showing a left hemi-scrotal mass post-operatively which actually was a haematoma but was thought to look like a persistent adenomatoid tumour [radiology report]; this mass resolved completely and was eventually diagnosed as haematoma. (There is no clear cut way of differentiating organised intra-scrotal haematoma from adenomatoid tumour radiologically on ultrasound scan).

Illustration 6

Illustration 6: Subsequent ultrasound scan 11 months post-operatively which had shown complete resolution of the haematoma.
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