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Paratesticular Leiomyoma – Case Report of A Rare Scrotal Mass.

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Abstract

Smooth muscle cells give rise to benign mesenchymal tumours known as leiomyomas. Although leiomyomas from the uterus are very common, leiomyomas from the scrotum are extraordinarily rare. They are usually solitary and can develop from scrotal wall, epididymis, spermatic cord or tunica albuginea. We present a case of single scrotal leiomyoma in a 55-year-old man who had a 1.5x1.8 cm, firm, slowly growing lump in his left scrotum. The mass was painless and firm.

An ultrasound scan of the scrotum indicated the presence of a heterogeneously hypoechoic lesion with no demonstrable vascularity abutting the tail of epididymis for which he underwent contrast enhanced MRI study of the scrotum for better characterisation of the mass. Histopathology results following excision revealed findings compatible with leiomyoma. A final diagnosis of Paratesticular leiomyoma was made. Here we report the clinical, radiological, histopathological and immunohistochemistry features of paratesticular leiomyoma.

Introduction

Anatomically, the scrotal lesions are divided into intratesticular and paratesticular pathologies. The epididymis, spermatic cord, vestigial remnants and tunica vaginalis make up the paratesticular area. (1)

The benign paratesticular tumours include lipomas, adenomas, leiomyomas, fibromas, hemangiomas, neurofibromas, and cystadenomas (2). Only 7% to 10% of all intra scrotal tumours are primary paratesticular tumours, making them uncommon. More than 75% of these lesions in adulthood develop from the spermatic cord (3).

Patients of all ages are affected by primary solid neoplasms of the paratesticular tissues, which are clinically significant. Majority cases present with a

slowly growing, non-tender tumour and are asymptomatic. Patients typically report of an expanding, non-tender lump. The most frequent epididymal tumour is an adenomatoid tumour, which is followed by a leiomyoma. Sarcomas must be taken into account in the differential diagnosis of all solid scrotal tumours in adult patients. (1)

Leiomyoma is a smooth muscle-derived tumour that can originate in many organs other than uterus and adnexae (4) including the spermatic cord, epididymis, renal pelvis,ureter, prostate gland, scrotum, and glans penis. (5) Only five occurrences of urinary tract leiomyoma have been documented in Southeast Asia, which is an extremely unusual site for this tumour (6). In the real world, the prevalence of paratesticular leiomyomas reached up to 17.7% (7). Primary ovarian leiomyomas (8), testicular leiomyomas (9), renal pelvis and ureteric leiomyomas (10), and other rare occurrences of leiomyomas have also been documented.

Case report

A 55-year-old male patient reported to the OPD with chief complaints of painless scrotal swelling on the left side. Patient gave a medical history of hypertension. The patient first noticed the swelling 1 year back and it gradually increased to the current size. The mass was palpable and was associated with occasional low-grade pain. There was no history of fever or trauma associated with it.

During physical examination, an ovoid swelling of size 1.5x1.8 cm was palpable in the inferior pole of left hemiscrotum appearing to be in continuity with the tail of epididymis.

The consistency was firm with bosselated surface and well-defined borders. The swelling is separately felt from the testis, is mobile and can get above the swelling. A normal testicle was discovered on the right side. The serum tumour markers hCG, lactate dehydrogenase, and alpha-fetoprotein were within the normal range.

Scrotal ultrasound examination indicated the presence of a heterogeneously hypoechoic lesion with no demonstrable vascularity abutting the tail of epididymis (Figure 1 and Figure 2).

In order to further characterize the paratesticular mass Contrast enhanced MRI was advised which revealed a well-defined altered signal intensity solid lesion measuring 1.6 x 1.3 x 1.3 cm (AP x TR x CC) present within the left hemiscrotum inferior to the left testis. It showed T1W isointense and T2W hypointense signal with no evidence of diffusion restriction/ blooming on SWAN sequence.

It was found to be in close association with the tail of the left epididymis. (Figures 3, 4, 5 and 6) On post contrast sequence, mild enhancement was observed (Figure 7). The remaining portion of the left epididymis was normal. Minimal free fluid was noted in bilateral hemiscrotum. (L>R). All the above features were highly suggestive of a benign paratesticular neoplasm most likely to be an adenomatoid tumour.

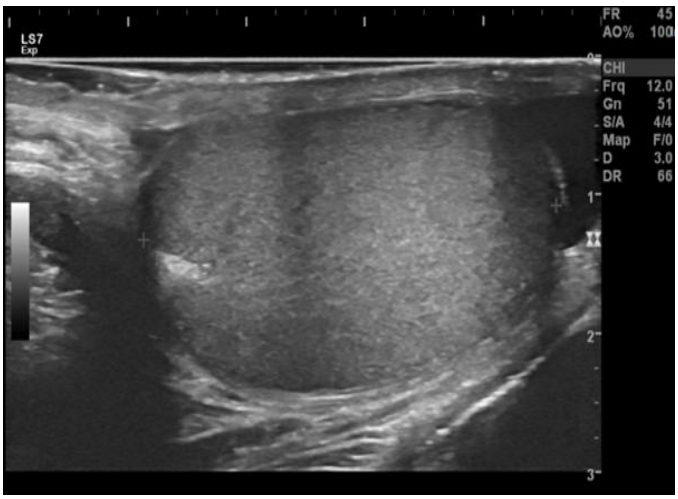


Figure 1: Ultrasound of the scrotum: Left testis appears normal. No evidence of focal lesions.

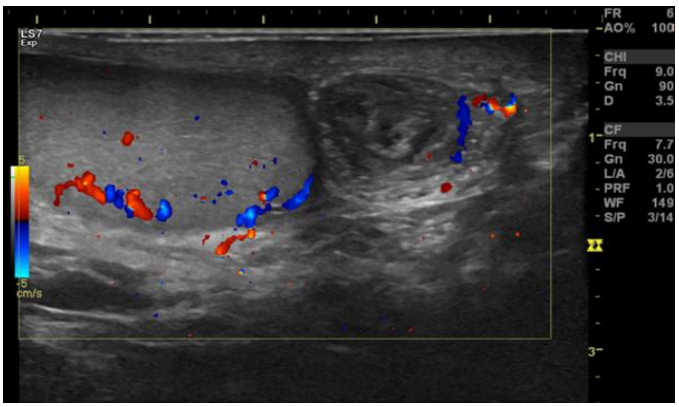
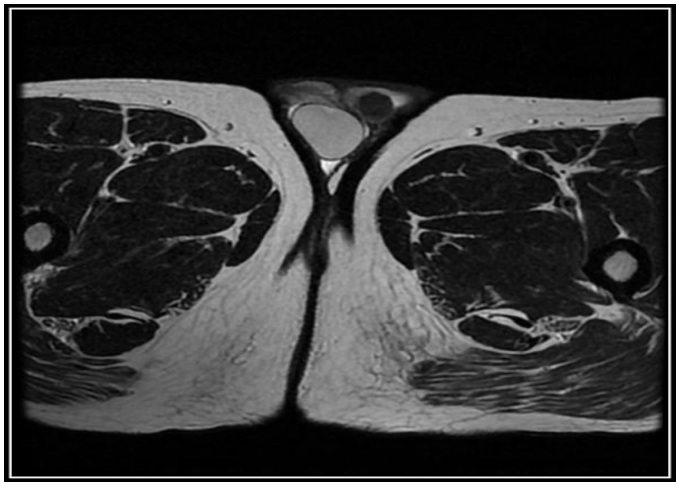
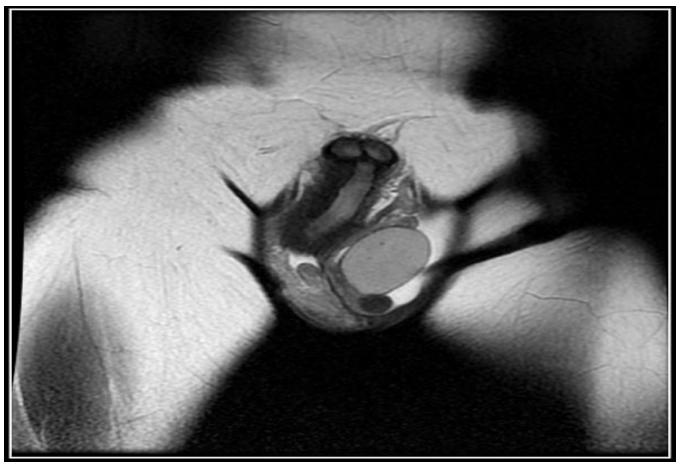


Figure 2: Color doppler ultrasound of the scrotum: Heterogeneously hypoechoic lesion with no demonstrable vascularity abutting the tail of epididymis.





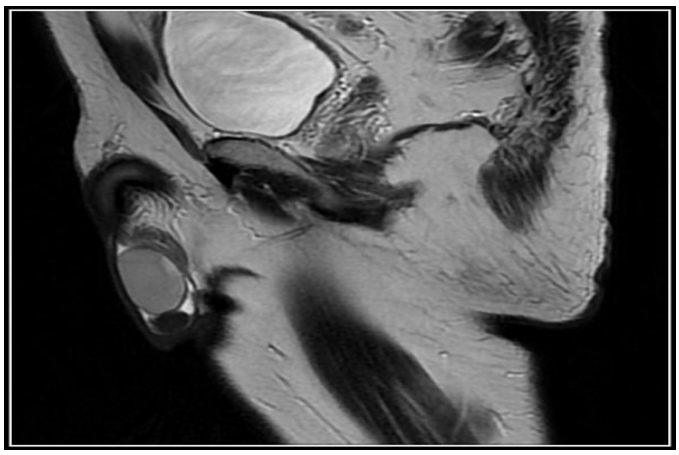


Figure 3, Figure 4, Figure 5: T2W MRI in all 3 planes Axial, sagittal and coronal shows a well-defined T2W hypointense lesion within the left hemiscrotum inferior to the left testis in close relation to the tail of the left epididymis

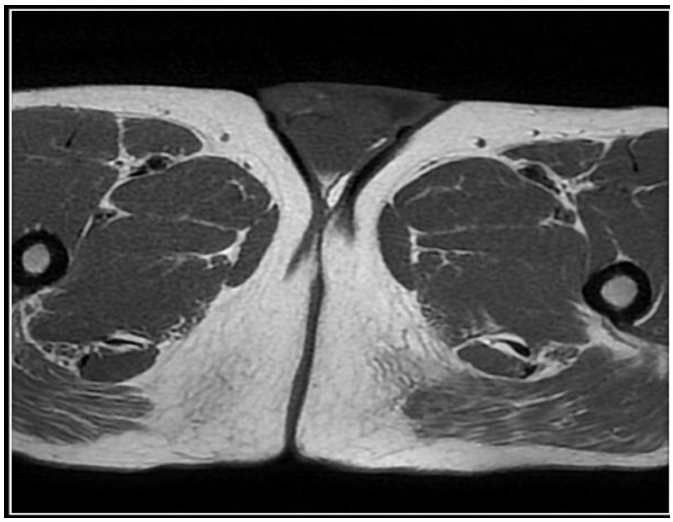


Figure 6: Ax T1WI shows a well-defined T1W isointense lesion within the left hemiscrotum inferior to the left testis in close relation to the tail of the left epididymis

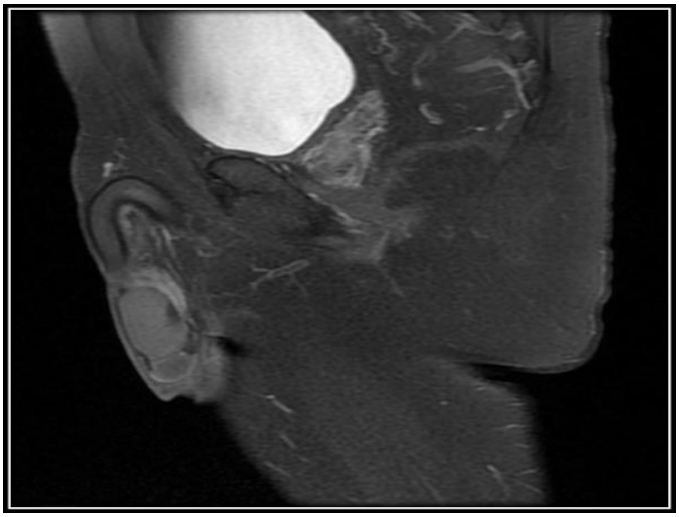


Figure 7: Post contrast Sag T1FS+C shows mildly enhancing altered signal intensity solid lesion within the left hemiscrotum inferior to the left testis in close relation to the tail of the left epididymis.

The testicle was then surgically explored through the inguinal area. The lump was determined to have originated outside of the testicle, in the paratesticular area.The mass was seen arising from the tail of epididymis and had close proximity with the testis. Around 10 ml hydrocele fluid was present when tunica vaginalis was opened. However, a high inguinal orchidectomy was done because it was difficult to separate the tumour from the testicle and there was a remote possibility of testicular malignancy.

The gross specimen measured 10 x 5 x 3.5 mms with the testes measuring 4 x 3 x 2.8 CMS. The attached cord was 6 cm long. The cut surface of the tumour revealed a grey white solid mass with whorling pattern measuring 1.5 x 1.4 x 1 cm.The postoperative course went without any complication, and the patient was stable enough to be discharged on the very first postoperative day. The excised mass was sent for histological analysis for additional evaluation.

A well-defined neoplasm made up of cells organised in overlapping fascicles and bundles was found by histological analysis. Each cell was spindle-shaped, had a lot of eosinophilic cytoplasm, and had a large vesicular nucleus. (Figure 8 and 9) No indication of abnormal cells or cancer was found. The diagnosis of paratesticular leiomyoma was confirmed by the existence of a mesenchymal spindle tumour on histology.On close monitoring of the patient for the past two months, no issues have been noted.

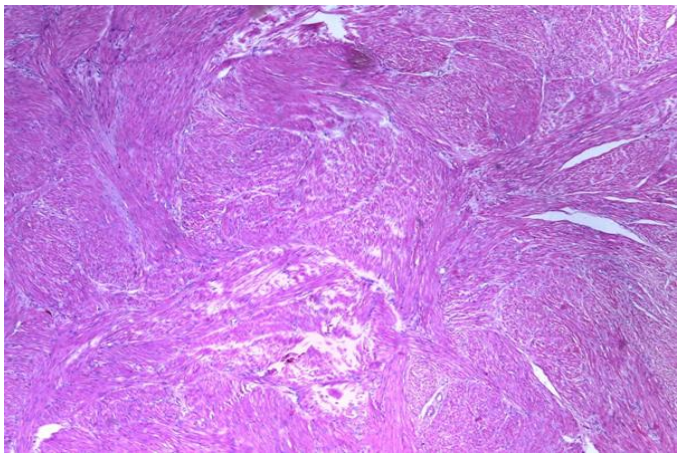


Figure 8: Microscopic Sections from tumour show a fairly well circumscribed neoplasm composed of cells arranged as interlacing fascicles and bundles.

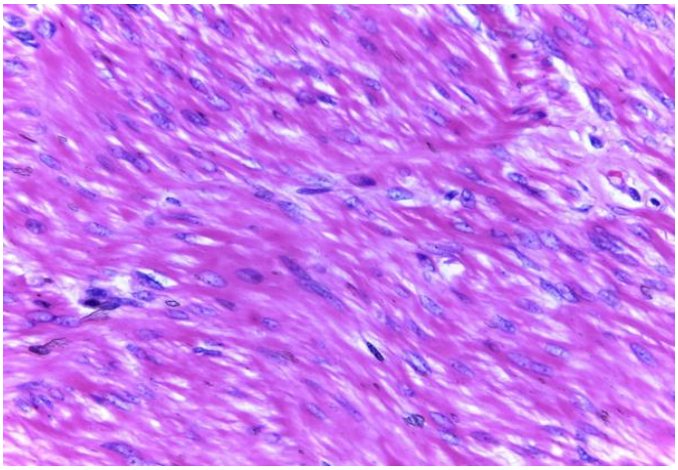


Figure 9: Microscopic sections: Individual cells are spindle shaped with abundant eosinophilic cytoplasm and plump vesicular nuclei.

Discussion

Patients of all ages can develop paratesticular tumours, which are rare but significant clinical lesions; the likelihood that they are benign is high—about 3% of cases are malignant (1,11,12). The paratesticular area is a complex anatomical region made up of the tunica layer of the spermatic cord, the epididymis, and the testicular

appendices. Mesenchymal, epithelial, or mesothelial cells can give rise to tumours in the paratesticular area. Only a few cases of leiomyomas in the paratesticular area have been documented (13).

Adenomatoid tumours, lipomas, leiomyomas, fibromas, hemangiomas, neurofibromas, and papillary cyst adenomas make up the bulk of benign paratesticular lesions in the scrotum. On the contrary, malignant tumours are uncommon and include malignant schwannoma and malignant fibrous histiocytoma, as well as liposarcoma, rhabdomyosarcoma, lymphoma, and fibrosarcoma. The majority of patients come with an indolent, slowly developing mass without any symptoms. Unfortunately, many solid extra-testicular masses have US features that are frequently non-specific, making a definitive diagnosis difficult to make in most situations. (1)

Smooth muscle is the source of leiomyoma, which is why it can be seen in many organs. According to the origin site, there are three different types of leiomyomas that can be distinguished with regard to the male genitourinary system: (1) leiomyomas that originate from the erector pili muscle (piloleiomyoma); (2) leiomyomas that originate from the smooth muscles of blood vessels (angioleiomyoma); and (3) genital leiomyomas (e.g., from the smooth muscles of the scrotum.(14,15) Although the renal capsule is where the bulk of male genitourinary tract leiomyomas are discovered, these tumours have also been seen in the tunica albuginea, spermatic cord, and epididymis(1,16).

Leiomyoma is a slow-growing, typically indolent tumour that can afflict people of all ages. It is more common in people in their fourth and fifth decades. (17) In our situation, the patient was a male who was 55 years old, which is a little older than the average prevalence. An earlier investigation identified a case of paratesticular leiomyoma in a 13-year-old child, which was one of the youngest cases ever identified. (18)

A non-tender lump in the scrotum is the most typical presentation of paratesticular leiomyoma. Pain in the inguinal region or the scrotum above the testicles may accompany this tumour. Most people delay to seek therapy when a tumour is asymptomatic and slowly expanding, until it becomes quite large, causing undesirable cosmetic effects or producing excruciating pain in the mass (19).

An ultrasound is often performed 7-8 years after mass manifestation (20). Adenomatoid tumour and fibroma show as firm, hypoechoic, or heterogeneous masses that may or may not display shadowing calcification. (21). However, research by Cassidy et al. found that MRI is the more precise and sensitive imaging modality for locating leiomyomas (22) In fact, MRI enables tissue characterisation because its signal intensity qualities enable the detection of fat, blood products, granulomatous tissue, and fibrosis. The following are typical MRI characteristics of leiomyoma: [1] Less contrast enhancement compared to nearby organs,[2] isointense signal on T1-weighted imaging, and [3] low signal on T2-weighted imaging. (22).

An accurate diagnosis can only be made by surgery. Paratesticular leiomyoma should be examined through the inguinal route and requires significant spermatic cord, testicular, and high cord ligation resection. (23) Our case underwent a testicular exploration via the inguinal route. Interestingly, a paratesticular tumour that was later determined to be leiomyoma was discovered.

Surgical excision is the primary mode of treatment for leiomyomas since they are often well-defined masses with a white or grey capsule. If there is suspicion of testicular cancer or if the tumour cannot be separated from the testis, a radical orchiectomy may also be performed (2,13). The lump was adherant to the testicle

in this case, raising the remote possibility of testicular cancer. In this case, a radical orchiectomy was unavoidable because it was difficult to delineate the boundary between the tumour and testicle.

When leiomyomas are examined histopathologically, mature smooth cells may be seen grouped in fascicles and separated from one another by collagenized stroma, but increased mitotic activity and the coagulation process may not be present. Under a microscope, the tumour seems to be made up of smooth muscle spindle cells that are grouped in interlacing bundles with various quantities of fibrous and hyalinized connective tissue (1). These tumours are divided pathologically based on four criteria: i) a maximum diameter of >5 cm; (ii) infiltrating margin; (iii) >5mitotic figures per high-power field; and (iv)substantial cytological atypia. An atypical leiomyoma is a tumour that meets two of the aforementioned criteria, an atypical leiomyoma is a tumour that meets three to four of the aforementioned criteria, and a leiomyosarcoma is a tumour that meets three to four of the aforementioned criteria. (24) The existence of a mesenchymal spindle tumour in this instance supported the paratesticular leiomyoma diagnosis.

An intraoperative frozen section may be done to rule out cancers after the bulk lesion has been removed. To determine the diagnosis of leiomyoma, immunohistochemical staining for SMA, caldesmon, and desmin must be positive. An inflammatory myofibroblastic tumour (IMT) of the spermatic cord is linked to desmin deficiency (25). This test is crucial to exclude leiomyosarcoma from the differential diagnosis since mouse double minute 2 (MDM2) amplification is the distinguishing feature of leiomyosarcoma. However, due to a lack of resources, immunohistochemistry was not an option in our case. Monitoring is crucial, and any recurrence needs to be recorded in order to completely rule out the possibility of malignancy.

Table 1: Comparison with other case reports

Reference	Age at diagnosis	Size of lesion
Current study	55 years	1.5x1.8cm
Irsayanto et al (13)	36 years	15 × 10 cm
Mardi K (5)	37 years	6 cm × 2.5 cm
Giriyan et al (26)	52 years	10x8cm

Conclusion

In summary, paratesticular leiomyoma is a very rare tumour that typically has an insidious development and non-invasive pattern. Most individuals would not require an orchiectomy if they receive prompt diagnosis and effective therapy. Because of its widespread availability, cost-effectiveness, and excellent sensitivity for detecting paratesticular masses, US is unquestionably the modality of choice for initial examination of scrotal pathologic disorders. However, MRI can further help with tissue characterization. (2).Final diagnosis requires histopathology and immunochemistry.

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