Asynchronous bilateral leiomyomata of tunica vaginalis: A case report

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Abstract

Leiomyomata are smooth muscle tumours that are slow growing and benign. We report a case of bilateral leiomyomata arising from tunica vaginalis. A 65 year old presented with a 5-year history of a slow growing left testicular mass and underwent radical orchidectomy. He presented again 18 months later with a slow growing right testicular mass and underwent local surgical resection. In both the cases the diagnosis of tunica vaginalis leiomyoma was determined through subsequent histopathological analysis. We hope to inform urologists to be aware of this benign rare entity as it can be cured through a simple, organ preserving surgical excision.

Introduction

Paratesticular leiomyomata are a rare intrascrotal entity derived from mesenchymal cells contained within the epididymis, the spermatic cord, the tunicae and testicular parenchyma. They are well-defined intrascrotal tumours often with a surrounding fibrous capsule. The tumour is solitary, painless and grows slowly over time. They are often mistaken for testicular malignancy leading to surgical excision.

To our knowledge this is the first case to describe asynchronous bilateral leiomyomata of the tunica vaginalis.

Case report

A 65-year old Caucasian male presented to the general urology clinic with left hemi-scrotum swelling. The swelling was first noticed five years ago on self-examination. Previously this had been investigated with an ultrasound scan which showed it to be 2.3 x 2.3 x 1.9 cm well defined heterogeneous mass, thought to be inflammatory change of epididymis, post trauma, or sperm granuloma.

During the following five years it had slowly grown over time, however the patient remained otherwise asymptomatic. The patient was re-referred due to discomfort. On examination, the patient had large painless mass, fixed to the left testicle. Trans-illumination test was negative. His right testis was of normal size with no associated scrotal swelling. There was no palpable inguinal lymphadenopathy or abdominal masses.

His bloods were unremarkable. Tumour markers for testicular cancer (α-fetoprotein, lactate dehydrogenase and β-human chorionic gonadotropin) were within normal range. Testicular ultrasound scan was repeated. The scan confirmed the presence of 5.5 x 4.5cm solid heterogeneous lesion with internal vascularity (Fig. 1a). It appeared to be separate from the testes, and likely arising from epididymis. The right testes and epididymis were normal. A pre-operative CT scan of the chest, abdomen and pelvis was performed and no abnormalities were detected.

The patient was counselled and consented for surgical excision of the mass and left testicle. He underwent radial left orchidectomy using the inguinal approach. His operation was performed as day case with no post-operative complications. Histological examination of the surgical specimen showed a well-circumscribed lesion 7 cm in diameter with a whorled cream cut surface, abutting native smooth of tunica vaginalis. Morphologically it comprised fascicles of unremarkable smooth muscle with no cytologic atypia, necrosis or mitotic activity.

Eighteen months later, patient was seen again in the general urology clinic with a three-month history of palpable mass in the right testicle. On examination, there was an oval shaped, firm painless lesion, separate from the testes. His bloods and tumour markers were normal. An Ultrasound scan of the right testicle was performed. USS described the lesion as a 5.8 x 4.6 cm mixed reflectivity mass with internal vascularity, identical to the mass in the previous scans (Fig. 1b).

The patient was advised and consented for local surgical excision of the right paratesticular tumour. The procedure was conducted via the inguinal approach. The right testis and cord were preserved.

Macroscopic examination again showed a macroscopically solid lesion with cream whorled cut surface, measuring 5.5 x 5.5 x 5.0 cm (Fig. 2). Microscopically it also showed a non-encapsulated well-circumscribed tumour comprising fascicles of bland smooth muscle with no worrying features(Fig. 3).

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Benign tumours of mesenchymal origin are rare in the genitourinary tract, the most common among them being leiomyomata.\(^1\) Leiomyomas are usually painless, indolent and slow growing.\(^2\) Within the scrotum, they can arise from the epidermis, spermatic cord, tunica albuginea, epidermis, spermatic cord, tunica albuginea, etc.\(^3\)

**Discussion**

Benign tumours of mesenchymal origin are rare in the genitourinary tract, the most common among them being leiomyomata.\(^1\) Leiomyomas are usually painless, indolent and slow growing.\(^2\) Within the scrotum, they can arise from the epidermis, spermatic cord, tunica albuginea, epidermis, spermatic cord, tunica albuginea, etc.\(^3\)

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**Fig. 1.** 1a. USS right hemi-scrotum 1b. USS left hemi-scrotum.

**Fig. 2.** 2a. Gross specimen of the lesion in situ 2b. Gross specimen of the lesion ex vivo.

**Fig. 3.** 3a. H&E section, 40x magnification, showing relationship to Tunica Vaginalis 3b. H&E section, 400x magnification, showing fascicles of benign smooth muscle.
tunica vaginalis, tunica dartos and testicular parenchyma.\textsuperscript{1}

To our knowledge only two other cases have been reported of leiomyomata arising from the tunica vaginalis.\textsuperscript{3,4} All the patients presented in their sixties, with a non-tender testicular mass during examination with the size ranging from 2.3 cm to 7 cm. Histologically, the tumour was seen as a well demarcated, grey-white capsulated lesion exhibiting a whorled appearance on cut surface. Microscopically, it consists of smooth muscle spindle cells in fascicular fashion with varying admixtures of fibrous and hyalinized connective tissue.\textsuperscript{3,4}

All the patients underwent orchidectomy, as malignancy could not be ruled out. While the majority of extra-testicular solid masses are benign (e.g. lipoma, adenomatoid tumours and leiomyoma), there is a three percent incidence of malignancy (e.g. liposarcoma, rhabdomyosarcoma and lymphoma).\textsuperscript{5} Sonography can be useful in describing location, composition (cystic or solid) and vascularisation. However this can be non-specific with variable findings, and thus not allowing a diagnosis of certainty.\textsuperscript{2} MRI can give better tissue characterisation and on table frozen sections would be ideal in avoiding a radical orchidectomy.

Patients who represent with contralateral masses with ultrasound showing similar characteristics to previous leiomyoma should be carefully examined. Pre-operative MRI scan is useful to procure diagnostic information. Local surgical resection via the inguinal approach should be offered to the patient with on table frozen sections if required. This allows the patient to avoid unnecessary radical orchidectomy and lifelong testosterone replacement.

Conclusion

This is a very unusual and interesting case of asynchronous bilateral paratesticular leiomyomata, apparently arising from tunica vaginalis. As far as we know no other case reports have described this. It highlights the fact that leiomyomas can re-present and be managed with a local surgical resection.

Consent

Written informed consent was gained for this case report and pictures, and is available for review.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.eucr.2019.101067.

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