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CASE REPORT

A Case of Benign Leiomyoma Arising from Dartos Muscle:An Unusual Form of Chronic Scrotal Swelling

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Abstract

Leiomyomas are benign mesenchymal tumors which arise from smooth muscle cells. They are most commonly encountered in the uterus but leiomyomas originating from the scrotum is a rare entity and very few cases are reported in literature so far. There are three types of superficial leiomyoma based on their site of origin: piloleiomyoma arising from arrector pili, scrotal leiomyoma arising from dartos muscle, and angioleiomyoma arising from smooth muscles of blood vessels. Herein, we report a case of scrotal leiomyoma in a 60 years old male which was clinically diagnosed as sebaceous adenoma and was excised. Histopathological and Immunohistochemical features were consistent with leiomyoma and final diagnosis of scrotal leiomyoma was given.

Keywords:

Scrotal Swelling, Leiomyoma, Smooth Muscle Actin

Introduction

Leiomyomas are benign mesenchymal tumors which arise from smooth muscle cells. They were first described in the uterus, by Virchow in 1854 as tuberculum dolorosum[1]. The uterus is known to be the commonest site of the tumor in women of reproductive age group. Extrauterine leiomyomas are classified based on their site of origin as piloleiomyoma (derived from arrector pili muscle of hair follicles) which is the most common type, angioleiomyoma (from smooth muscle of vessel wall), or genital leiomyoma (from dartos muscle of scrotum, nipple, vulva) which is a rare entity[2].

The first case of scrotal leiomyoma was described in 1858 by Forsters and its prevalence rate is reported to be 1 of 1000 of all scrotal tumors by a review of 11,000 scrotal tumor cases [3]. It can arise from the epididymis, spermatic cord, tunica albuginea, or scrotal wall. Exact cause of development of this rare subtype is unknown, but may be related to male hormone androgens, as studies have demonstrated presence of androgen receptor expression in these tumors[1]. They are further categorized into conventional or typical leiomyoma and atypical or symplastic leiomyoma that have bizarre nuclei which may mimic malignancy, and leiomyosarcoma[4]. The tumor is almost always solitary and grows slowly if not excised. Keeping in view the rarity of this tumor, we report a case of solitary scrotal typical leiomyoma.

Case Report

A 60-years-old man presented with the complaints of a painless mass in the right side of scrotum since 5 years

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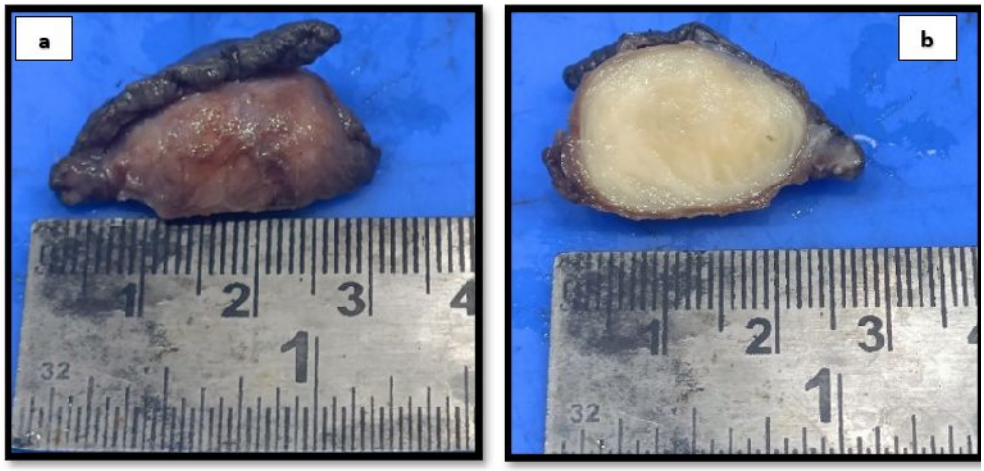

Fig. 1 (a) Excised Skin Covered Soft Tissue Mass; (b) Cut Section of the Tumor Showing Solid, Grey White, Homogenous Lesion With Whorled Appearance.

which was insidious in onset. There was no history of sudden increase in size of the swelling, fever, or trauma. Physical examination revealed a single well-defined, soft to firm, nontender, mobile lump of 3× 2 cm on anterior aspect of right scrotum. It had no palpable connection to the testis, epididymis or spermatic cord. The overlying skin was normal without any ulceration. Both the testes were normal without any palpable inguinal lymph nodes. Routine blood and urine laboratory tests were normal. Ultrasound scan of the scrotum showed a well-circumscribed mass with mixed echogenicity predominately hypoechoic measuring 2 x 1.5 cm with no internal vascularity seen inferior to and clearly separate from the adjacent right testicle. No necrosis was observed within the mass. The swelling was excised with a provisional clinical diagnosis of sebaceous adenoma and sent for histopathological examination.

Gross examination showed a single, skin covered, firm tissue mass measuring 3 × 1.5 × 1 cm. Cut surface showed solid, grey white, homogenous lesion with whorled appearance (*Fig 1*). Haematoxylin and eosin stained microsections shows keratinized stratified squamous epithelium with a underlying well-circumscribed tumor composed of spindle shaped cells having blunt ended elongated nuclei and eosinophilic cytoplasm arranged in interlacing fascicles and bundles admixed with varying amounts of collagen. No nuclear pleomorphism, mitoses or necrosis noted in the sections studied. Periphery of the lesion shows bundles of smooth muscle layer (*Fig 2*). Immunostaining for smooth muscle actin (SMA) was done on paraffin embedded sections. The tumor showed SMA positivity in 100% of the cells, confirming smooth muscle phenotype (*Fig 3*). On the basis of histopathological and immunohistochemical profile diagnosis of scrotal leiomyoma was made.

Discussion

Leiomyomas may originate from any anatomic location of smooth muscle in the genitourinary system. Only few cases of leiomyomas have been reported arising from the renal pelvis, bladder, spermatic cord, and epididymis, prostate as well as the glans penis[5]**.** Scrotal masses are primarily classified as intratesticular or extratesticular, and either solid or cystic. Solid intratesticular masses are malignant in 90%-95% of cases which includes germ cell and non–germ cell tumors, metastatic lesions, lymphoma, and leiomyosarcoma. Extratesticular masses are usually benign which includes lipoma, adenomatoid tumor, papillary cystadenoma, leiomyoma, fibrous pseudotumor, sarcoid granuloma, and polyorchidism. Most common extratesticular solid tumor is lipoma followed by adenomatoid tumor[6].

Scrotal leiomyoma is a rare tumor and its prevalence rate is reported to be 1 of 1000 of all scrotal tumors by a review of 11,000 scrotal tumor cases. It may arise from the epididymis, spermatic cord, tunica albuginea, or scrotal dartos muscle. They are more common in Caucasians between the fourth and sixth decades of life[3]. The most common presenting symptom is a non-tender mass in the

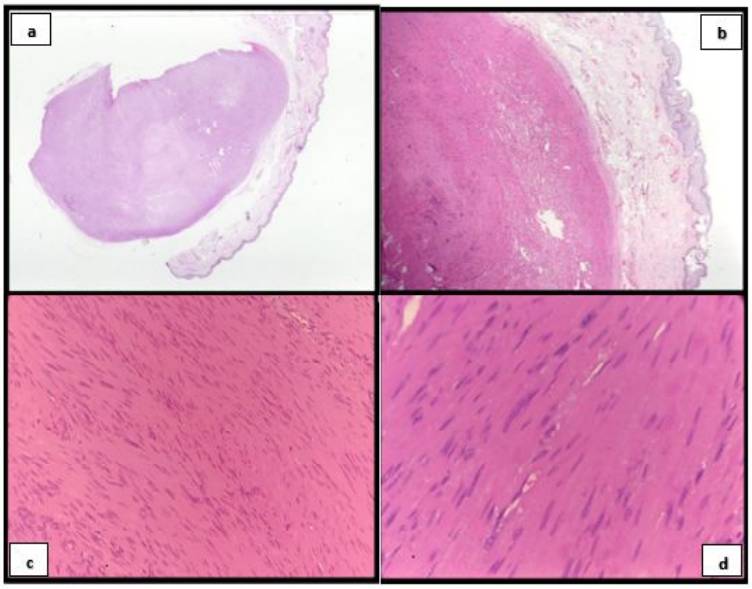

Fig. 2 (a) Whole mount View Shows Skin with underlying tumor, (b) x40 shows keratinised Stratified Squamous Epithelium with an Underlying well Circumscibed tumor; (c) x100 shows fascicles and interlacing bundles of spindle shaped smooth muscle celss; (d) x400 indivdual cells show moderate amount of eosinophilic cytoplasm, elongated blunt ended nuclei and Vesicular chromatin with inconspicuous Nucleoi

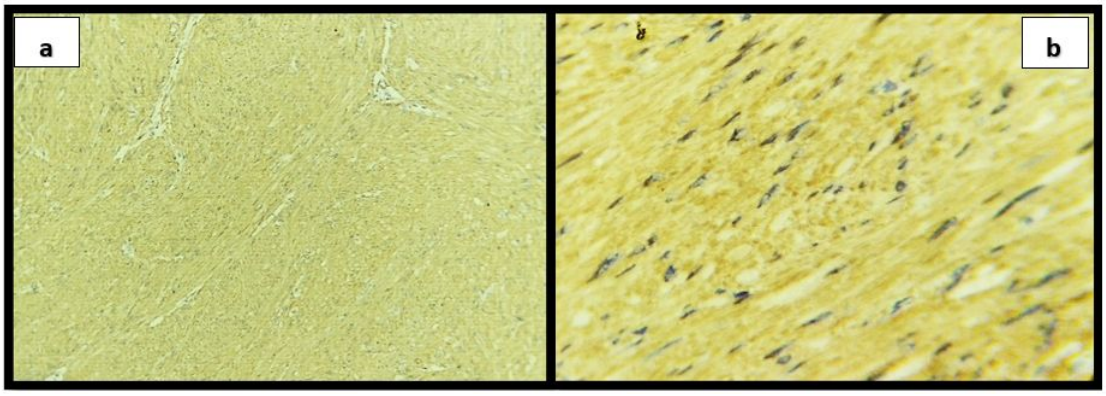

Fig 3 : (a) x100, (b) x400 Positive Expression of Smooth Muscle Actin (SMA)

scrotum and may be accompanied by pain in the scrotum above the testicle or in the inguinal area. When the tumor is asymptomatic or grows slowly, patients often delay seeking treatment until the lesion becomes large and cosmetically undesirable[7]. Clinically, the differential diagnosis includes a sebaceous cyst, fibroma and, if painful, a schwannoma could be considered. [5] Ultrasound is the first line of imaging in patients with known or suspected scrotal masses. It is a noninvasive and accurate way to evaluate the size, echotexture, and borders of the mass and helps in detecting any associated features such as calcifications or necrosis. It is also a reliable method to distinguish between intratesticular and extratesticular scrotal masses, with a 92%-98% sensitivity and 95%-99.8% specificity for testicular malignancy. [6]

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Leiomyomas are usually well-capsulated, solid grey white masses. Microscopically, the tumor consists of smooth muscle cells arranged in interlacing fascicles and bundles with varying admixtures of fibrous and hyalinized

connective tissue. In immunohistochemical examination; positive staining for smooth muscle actin, caldesmon and desmin is important to confirm the diagnosis of leiomyoma. S-100 negativity is necessary to exclude neurofibroma and schwannoma. Low mitotic activity (Ki67 proliferation index) is also compatible with leiomyoma. [8] Benign smooth muscle tumors of the scrotum are classifed either as typical or atypical leiomyoma. Four histologic criteria are used in this classifcation: [1] size ≥ 5 cm in widest diameter, [2] presence of infltrating margins, [3] ≥" 5 mitotic figures per 10 high-power felds and [4] moderate cytological atypia. Those with any one of these features are described as typical or conventional leiomyoma while tumors with two features are atypical or symplastic leiomyoma. The occurrence of three or more of these features renders such a leiomyosarcoma[9]. In the present case, the tumor was 2 cm in its greatest dimension, without cytological atypia, no mitoses per 10 HPF, and without infiltrating margins. Hence, a diagnosis of benign typical leiomyoma of scrotum was given. Surgery for even large lesions should be conservative if it is clear that it does not arise from the testis or adnexal structures but is of cutaneous origin; pathologic frozen section should be utilized to confirm the clinical suspicion if there is doubt regarding the extent of surgical excision. If frozen section suggests the diagnosis of leiomyoma, wide local excision is sufficient for management of such lesions[10].

Conclusion

Scrotal leiomyoma is a rare benign mesenchymal neoplasm, usually manifests as a painless lump in the scrotum. It is imperative to be considered as a differential diagnosis of paratesticular solid mass to provide treatment with testicular preservation, especially in young men. While it can be challenging to differentiate leiomyoma from other benign or malignant growths based on pre-operative imaging, it aids in assessing the invasion into the adjacent structures or the inner structure of the lesion. Nevertheless, surgical excision is sufficient for complete cure and histopathological examination will confirm the diagnosis.

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