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CASE REPORT

Scrotal Leiomyoma; an Unusual Cause for Scrotal Mass

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Abstract: Leiomyomas are the most common benign mesenchymal tumors arising from smooth muscle cells. Cutaneous leiomyoma constitutes small percentage of all the leiomyomas. However, genital leiomyomas, such as those of the scrotum have an even lower incidence rate. A 43-year-old man presented with complaints of a painless swelling in the left side of the scrotum. A clinical diagnosis of scrotal wall fibroma was made and the swelling was excised which had homogenous, grey-white areas with whorling and histologically showed features of leiomyoma. Here we present a rare cause of scrotal mass in a middle-aged man.

Keywords: spindle cell, actin, mitosis, smooth muscle

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1. Introduction

Leiomyomas are the most common benign mesenchymal tumors arising from smooth muscle cells. They are the most common benign tumor of the female genital tract in women of the reproductive age(1). The extra uterine locations of leiomyoma are scrotum, ovaries, bladder, lung, vascular structure and spermatic cord(1). Superficial leiomyoma of the skin and subcutaneous tissue is of basically three types based on the origin of tissue:piloleiomyoma(tumor of arrector pili muscle), angioleiomyoma(tumor of smooth muscle and blood vessels), and genital leiomyoma(from smooth muscle of the nipple, vulva and scrotum)(2). Cutaneous leiomyoma constitutes 5% of all the leiomyomas, however genital leiomyomas, such as those of the scrotum have an even lower incidence rate(3). The first case of the latter was accounted for by Forsters, in 1858(4).

Scrotal leiomyoma is an unusual variety which can originate from the scrotal wall, spermatic cord, epididymis, or tunica albuginea. Siegal and Gaffey described 11 cases of scrotal leiomyoma out of a total of 11, 000 scrotal tumors(5). We report this case to emphasize on the rarity of this tumor.

2. Case Report

A 43-year-old man presented with complaints of a painless swelling in the left side of the scrotum since eight years, which was single, well defined, soft to firm, non-tender,mobile, measuring 3x3x2.5cm on the anterior aspect of the left scrotum. It had no palpable connection to the testis, epididymis or spermatic cord. Bilateral testis and the overlying skin was normal. There was no inguinal lymphadenopathy. Radiological details were not available in this case. A clinical diagnosis of scrotal wall fibroma was made and the swelling was excised.

Gross examination showed a skin partially covered globular tissue measuring 3x3x2.5cm (figure1). Cut section showed homogenous, grey-white areas with whorling. Histopathological evaluation showed a well-circumscribed tumor in the sub-epithelium comprising benign spindle cells arranged in interlacing bundles and fascicles arranged in whorling pattern and admixed with collagen(figures 2 &3). Individual tumor cells were uniform in size, spindle-shaped with cigar-shaped nucleus having blunt ends and inconspicuous nucleoli,andmoderate amount of eosinophilic cytoplasm. No evidence of cytological atypia or mitoses were noted. No areas of necrosis were seen. Hence, a diagnosis of scrotal leiomyoma was made. As the morphology of the cells were classic the immunohistochemistry was not required.The patient was followed up for three years and he was disease-free and asymptomatic.

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3. Discussion

Genital leiomyomas are uncommon and often a poorly documented subset of pilar leiomyomas. They are comprised of smooth muscle neoplasms which have seemingly taken origin from the smooth muscles surrounding the pilosebaceous units which have been documented in the vulva, nipple,and the scrotum(6). Newman and Fletcher in their analysis found that compared with the leiomyomas of the nipple and cutaneous leiomyomas, which are quite similar in terms of size and histology, the vulva and scrotal leiomyomas are unique clinicopathologically(6). These are rare and have not been documented frequently in the literature. The latter tend to be larger and more circumscribed. Based on their experience, they recommended that because the vulva and scrotal tumors are distinct from other cutaneous smooth muscle neoplasms they should be categorized independently and not grouped with the former.

Scrotal leiomyomas are rare and usually arise from the dartos muscle. They are usually asymptomatic and commonly seen in middle-aged white men(5). They occur in the 4th to 6th decades of life and present as painless solitary, slow-growing cutaneous nodules. The asymptomatic, slow-growing and painless nature of this tumor could be the reason for late presentation of the patient in our case. The clinical features of scrotal leiomyoma maybe overlapping, hence the clinical differential diagnoses to be considered include fibroma, schwannoma and sebaceous cyst which can be readily differentiated on histomorphology. Any ulceration overlying the scrotal skin surface points towards the possibility of a squamous cell carcinoma (SCC). If there is any adhesion to the underlying testis, the possibility of atypical leiomyoma or leiomyosarcoma should be considered. The fibrous tissue with presence of pointed spindle cells having tapering ends indicates a likelihood of fibroma. In cases of schwannoma, which is a benign nerve sheath tumor, hyper- and hypocellular areas with Verocay body formation are seen which have a characteristic histomorphology. Owing to a paucity of cystic nature of leiomyomas, the sebaceous cyst can be ruled out during gross examination itself.

Radiologically, the extra testicular scrotal swellings are usually cystic in nature, while among the solid lesions the most frequent is lipoma(7). Though the radiological information in this case was not available, these benign adipocytic tumors are circumscribed, avascular and echogenic in nature. Adenomatoid tumor is second most common benign tumor of the scrotum. There can be benign proliferations secondary to trauma and/or infections which can result in fibrous pseudotumors. Scrotal leiomyomas are believed to be the most infrequent of solid scrotal masses. Ultrasonography (USG) is an easy and accurate method for evaluating scrotal masses. On USG, leiomyomas, Leiomyomas show well-defined, heterogeneously hypoechoic, vascular masses which are clearly demarcated from the surrounding tissue and may also show calcifications. Leiomyoma on T1-weighted image are isointense but show a low signal on T2-weighted imaging and show reduced enhancement compared to testis.

Histopathology remains the gold standard for the final diagnosis. Scrotal smooth muscle tumors, on histomorphology, show interlacing fascicles of benign spindle cells with blunt-ended cigar-shaped nuclei and moderate eosinophilic cytoplasm. These are further graded as benign, atypical, and malignant based on four pathological criteria: mitosis ≥ 5/10hpf, size of ≥5cm in greatest dimension, moderate cytological atypia and infiltrating margins.[6,8] Tumors with any one of the above-mentioned criteria are considered as benign while those with any two are atypical. Those fulfilling three to four of them are leiomyosarcomas. In complex cases, immunohistochemistry with an appropriate panel of markers can help in diagnosis. This should include smooth muscle actin (SMA), desmin and Ki-67 labelling index for confirmation.

Newman and Fletchermade a review and explained on mitotic activity as a potential criterion for malignancy. This necessitates the pathologists to search for mitosis meticulously.[6] Conventional and atypical leiomyoma behave similarly. They are managed by surgical excision with adequate margin in cases where atypia is present. However, leiomyosarcoma needs a wider resection(8). Radiation is avoided as it causes malignant transformation. Follow-up is needed to rule out malignancy in case of recurrence. Our patient is doing well after three years of surgery and he had no atypia, mitosis, or necrosis in the tumor. As is shown in this case, this report highlights the clinico-pathological features of scrotal leiomyoma and emphasizes that we must consider leiomyoma in the differential diagnosis of any scrotal swelling.

4. Appendix

4.1. Ethical consideration

The patient was contacted over phone and complete informed consent was obtained. He has given his complete consent but could not come for signing a written record because of the COVID-19 pandemic.

4.2. Conflict of interest

None.

4.3. Funding and support

None.

4.4. Author's contributions

All the authors have shared the same workload and thereby are entitled to equal contribution.

4.5. Acknowledgement

None.

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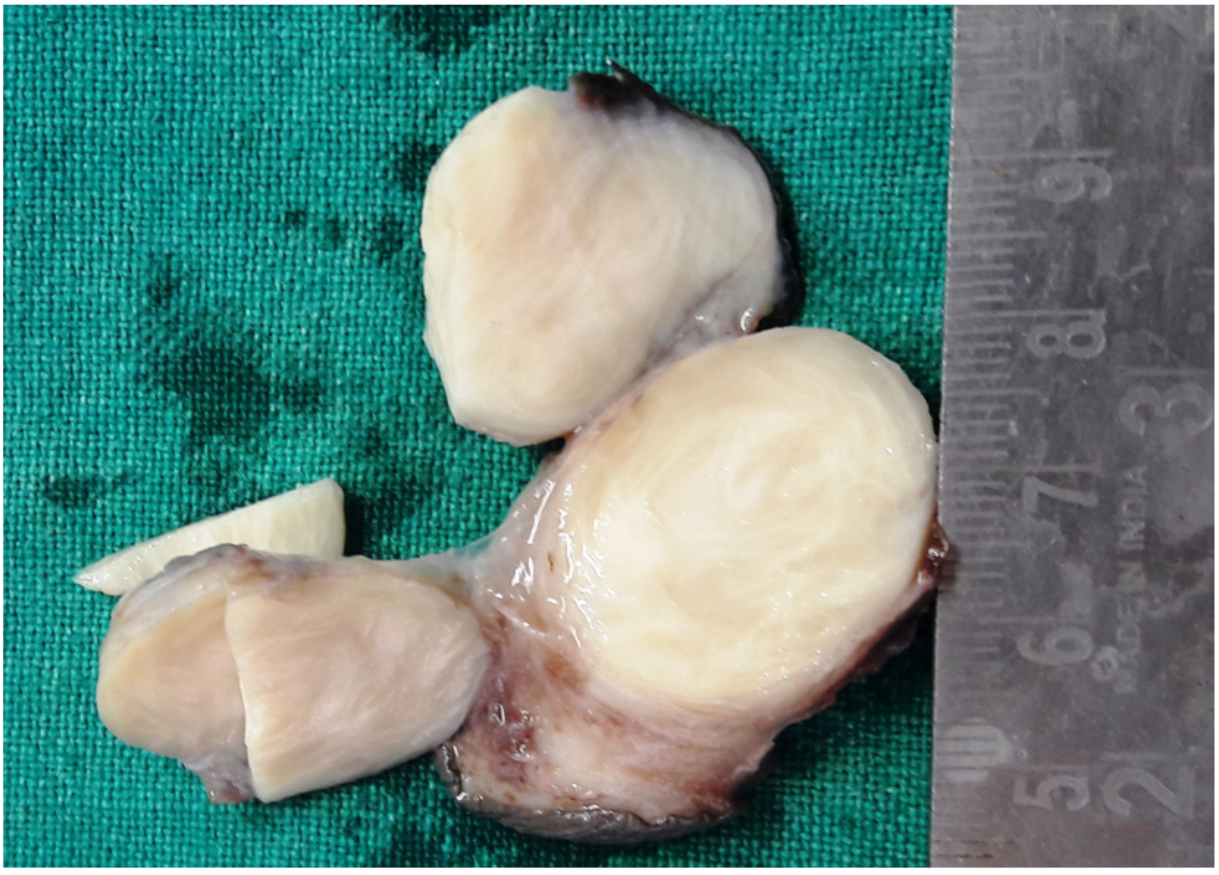

Figure 1: Gross specimen showing a globular tissue measuring 3x3x2.5cm. Cut section shows solid homogenous, grey-white firm areas with whorling.

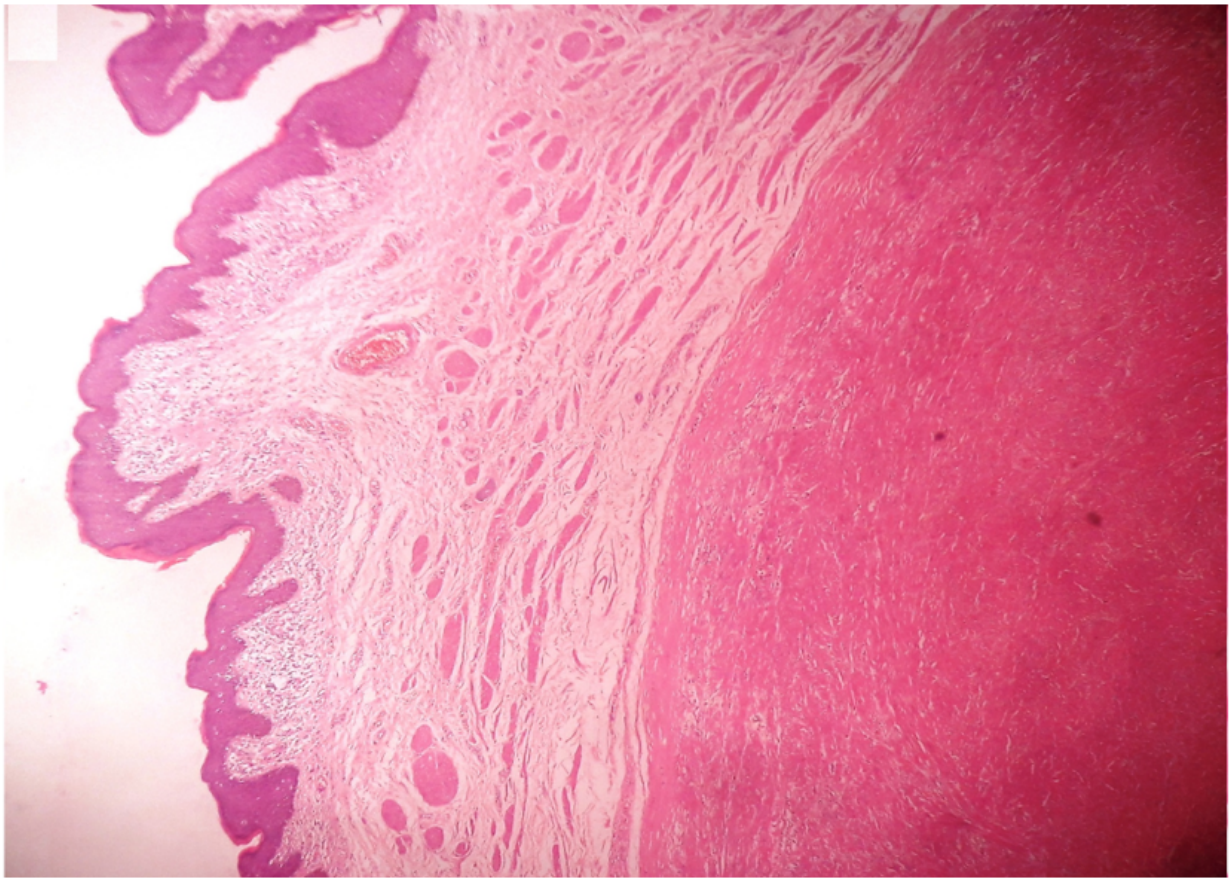

Figure 2: Section shows skin with a well-circumscribed tumor in the sub-epithelium (H&E, 40X).

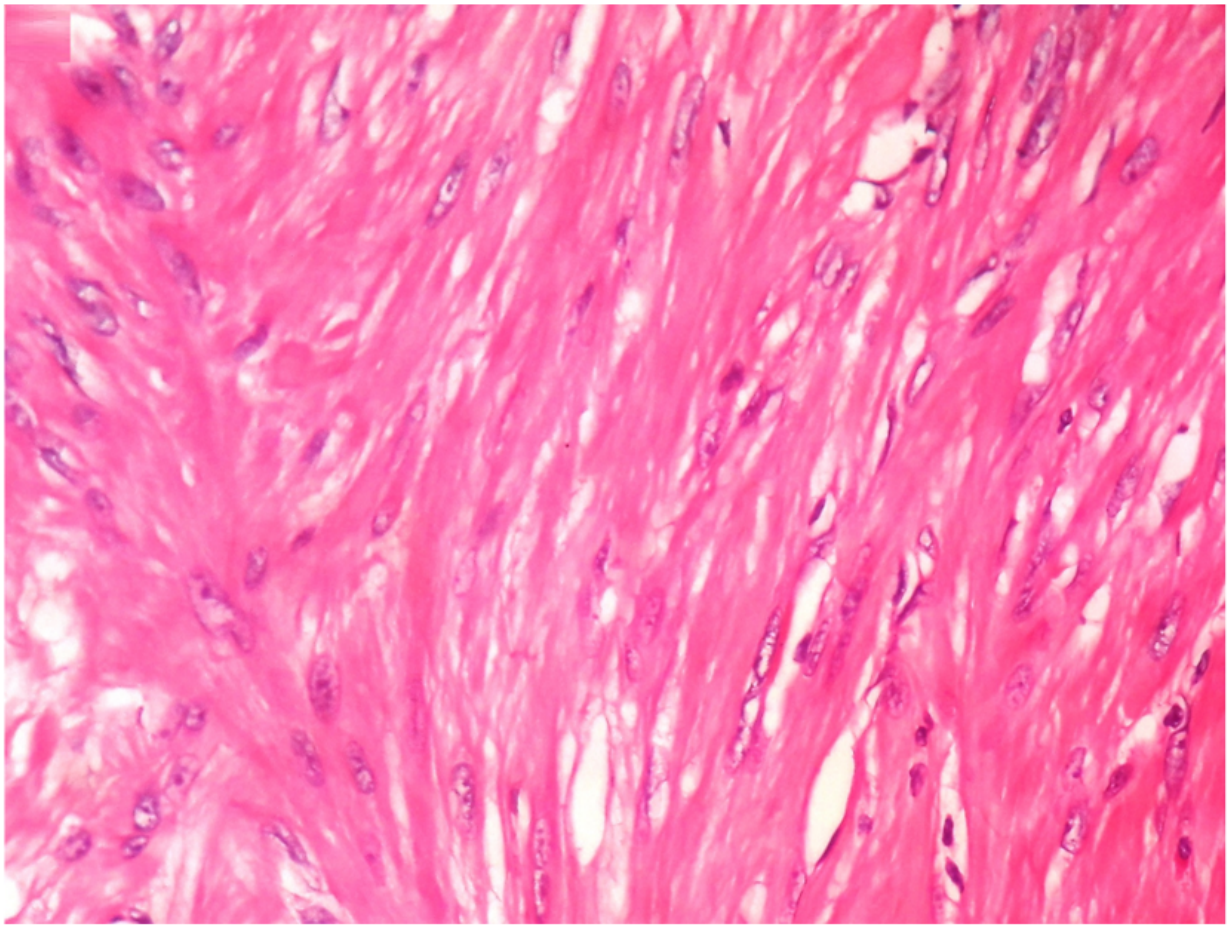

Figure 3: Section shows interlacing fascicles of spindle cells showing cigar-shaped nuclei and eosinophilic cytoplasm. No mitosis or necrosis is seen (H&E, 400X).