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Case Report

An Extremely Rare Case of Recurrent Scrotal (Dartoic) Leiomyoma

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ABSTRACT

Leiomyoma arising from the scrotum is a rare entity. The literature records less than 50 cases of scrotal leiomyomas, with one recording leiomyoma presenting with torsion. However, this is a case of a recurrence of a benign scrotal leiomyoma in a 17 year old patient, which is extremely rare, and probably the first of its kind. A similar lesion was excised from the same site and was a histopathologically-proved benign leiomyoma, which recurred again after a period of 2 years, achieving a size of 2.5 cms. It was excised, and was once again proved a benign leiomyoma, by histopathological features. The patient was followed for 5 months, during which it showed no recurrence. It is an extremely rare case, as it presented in young age, was painful and recurred.

Key words: Rare case report, leiomyoma scrotum, recurrent, dartoic leiomyomas.

INTRODUCTION

Leiomyomas are benign neoplasms that may arise from any structure or organ containing smooth muscle, commonest being the uterus. The majority of genitourinary leiomyomas are found in the renal capsule, but this tumor has also been reported in the epididymis, spermatic cord, and tunica albuginea. Leiomyoma arising from the scrotum is a rare entity. Here is a detailed case report of a very rare case of recurrent leiomyoma arising from tunica dartos, called dartoic leiomyoma.

CASE PRESENTATION

A 17 year old patient presented with a painful swelling over the right side of scrotum. The swelling was present since last 3 months, has gradually increased in size from <1 cm to 3cm (in the largest dimension), was still gradually increasing in size. It now measured 2*2*2 cm. It was visible as a separate swelling and at a higher level than the right testis. The swelling was firm, tender, without any raised temperature, non-fluctuant, non-transilluminant, having no cough impulse, having restricted mobility, indentation sign negative, traction test negative, present as a separate swelling from the underlying testis as well as from the overlying skin. A 2 cm well-healed linear scar was present over the swelling, suggesting the site of previous incision for a similar swelling and thus, a recurrence. There was no abnormality of the overlying skin, e.g., punctum, pus point or erythema. The swelling was painful. The pain was continuous and dull in nature and was present since the last 2 months. It had no aggravating or relieving factors. There were no palpable inguinal lymph nodes. There was no associated urinary tract infection, or a history of insect bite, mumps or any trauma. There was no significant personal or family history.

Past history

The patient was operated for a similar swelling at the same site 2 years back. The histopathological report showed a leiomyoma. However, the information regarding the tissue of origin was unavailable.

Radiological investigations

The ultrasound showed a well-defined heterogeneous and hypoechoic lesion of 24*22 mm at the root of right scrotum, possibility of arising from the spermatic cord internal vascularity was present with multiple foci of calcification.

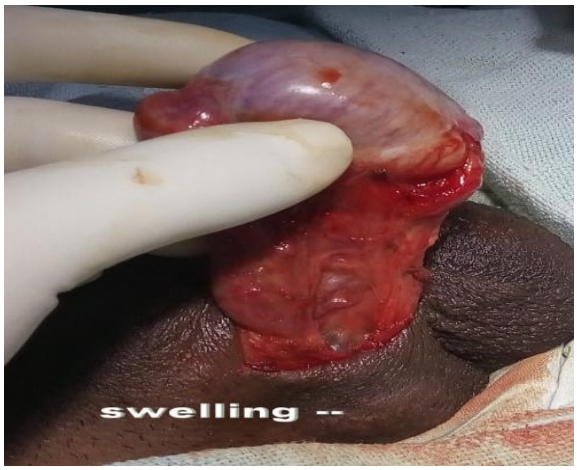

Fig. 1: Swelling can be seen distinct from the cord

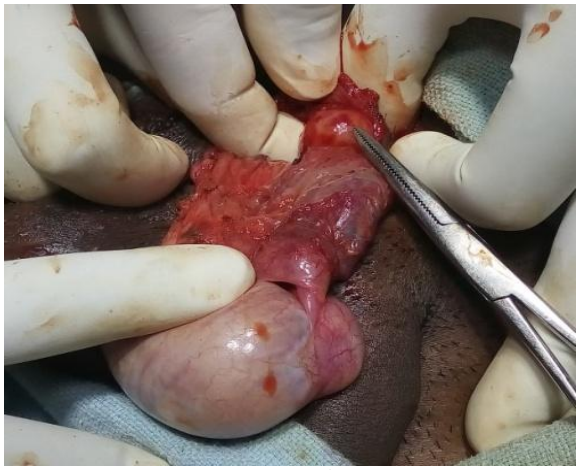

Fig. 2: Swelling is seen arising from the scrotal tissues

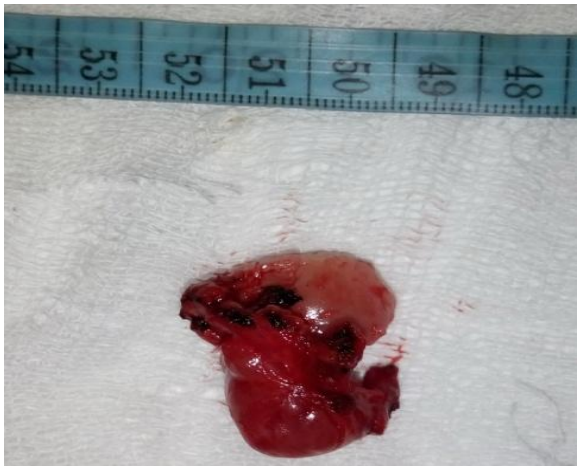

Fig. 3: The excised specimen

Intraoperative findings were a single firm whitish grey swelling which seemed to be arising from the spermatic cord; however, a distinct plane was obtained and the swelling actually was arising from the scrotal wall, mostly, dartos layer. It was excised and sent for further histopathological examination.

The histopathology report showed a well-defined mass lesion composed of interlacing fascicules of spindle cells having moderate amount of eosinophilic cytoplasm with central spindled nuclei and perinuclear vacuoles, all features suggestive of a leiomyoma.

Post-operatively, the patient was followed for 5 months, during which there was no recurrence.

DISCUSSION

The smooth muscle tumours of the scrotum originate from the subcutaneous tissue, which comprises of the dartos muscle, and are extremely rare neoplasms. [1] Among many such tumors, scrotal leiomyomas constitutes one rare group arising from the dartos muscle, with <50 cases reported. [1] Seigel and Gaffey demonstrated the rarity of this tumor by finding only 11 cases in a review of 11000 cases of scrotal tumor. [2]

Scrotal smooth muscle tumours are categorized as leiomyoma, atypical leiomyoma and leiomyosarcoma. Leiomyomas are benign smooth muscle tumors which have been found in many locations, including the epididymis (where it is the 2nd most common neoplasms comprising of 6% of all epididymal tumours), spermatic cord, tunica dartos, tunica albuginea and within the testicle. [3,4] However, they are rarely found arising from the scrotum. They usually present between the fourth and sixth decades of life. Grossly, they present as a circumscribed swelling or pedunculated scrotal mass. [5] Bilateral leiomyomas have been reported, but are very rare. [4] They are usually painless; however, pain and hydrocele may be an associated feature. [1,6] It usually presents

with painless solitary small cutaneous lesion measuring 1-14 cm with the average of 6.4 cm. [8] The slow growing nature of the tumor pushes the nerve trunk outwards, rather compressing it, hence they are painless. [1] Because of the asymptomatic, painless, and slow growing nature of the tumor, patients usually present late with an average of 7.6 years between the patient's recognition of the tumor and its surgical removal. [2]

Clinically, the differential diagnosis includes a sebaceous cyst, dermatofibroma, neurofibroma and schwannoma. Ultrasound evaluation is the imaging modality of choice for evaluating intra-scrotal pathology. The histopathological feature is the proliferation of tumor cells with characteristics of smooth muscle cells and act in positive immunohistochemistry.

Four pathological features are used to grade scrotal smooth muscle tumours that include

(i) size ≥5 cm in greatest dimension
(ii) infiltrating margin
(iii) ≥5 mitotic figures per 10 hpf
(iv) moderate cytological atypia

Tumors with only one of the above-mentioned features are considered benign. Those fulfilling two of the criteria are diagnosed as atypical leiomyomas. Tumours showing three to four of these criteria are leiomyosarcomas. [7] Special histochemical stains, e.g., Masson's trichrome, can demonstrate the myogenic nature of the tumour. Immunohistochemistry plays a vital role in determining the nature of spindle cells and conferring a final diagnosis. The smooth muscle tumour shows cytoplasmic positivity for smooth muscle actin and desmin and is negative for S100 protein.

A paucicellular tumour with pleomorphic atypical nuclei with a low mitotic activity is termed as pleomorphic/bizarre/symplasmic or atypical leiomyoma. Nuclear atypia in these tumours represents only a degenerative change. In every histologically diagnosed case of an atypical leiomyoma, the possibility of a leiomyosarcoma has to be ruled out. Definite criteria are quite useful to differentiate an atypical leiomyoma from a leiomyosarcoma at this region. It is important to recognize this entity mimicking a leiomyosarcoma. Newman and Fletcher studied a series of smooth muscle tumours localized to external genitalia, further suggesting a categorization of vulval and scrotal lesions separately as they are clinico-pathologically a distinct group of smooth muscle tumours. [8]

Typical leiomyomas and atypical leiomyomas behave in a similar fashion; hence they are managed with simple excision. However, leiomyosarcomas require a wider resection with negative margins. Radiation should be avoided as it may induce a malignant transformation. Previous reported cases on cutaneous atypical leiomyomas documented no recurrence or metastasis on a short follow-up period of 5 years. Because of the paucity of reported cases with short follow-up periods, a vigilant clinical surveillance of such patients is prudent.

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