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Genitourinary

Scrotal sac leiomyoma: Case report of a rare benign scrotal mass

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ABSTRACT

Leiomyomas are benign mesenchymal tumors, the overwhelming majority of which are located in the uterus. Rare cases arise in other organs, including the scrotum, pelvis, bladder, and spermatic cord. This report evaluates the case of a 37-year-old man with a history of prior left inguinal hernia repair, who presented with a painless right scrotal mass. He first noticed the mass approximately 1 year prior to his initial visit. Subsequent ultrasound of the scrotum demonstrated a 5-cm circumscribed, hypoechoic, mildly vascular extratesticular mass located within the right hemiscrotum. Based on the initial imaging, the differential diagnosis included lipoma, adenomatoid tumor, papillary cystadenoma, leiomyoma, fibrous pseudotumor, sarcoid granuloma, sarcoma (including liposarcoma, rhabdosarcoma, or leiomyosarcoma), lymphoma, and an extranumerary testis. The mass had circumscribed margins, suggesting an encapsulated lesion, and was completely separate from the testicle on ultrasound. Despite this, testicular malignancy was not entirely excluded as a diagnosis, although it was considered far less likely. The patient was definitively treated with surgical excision of the mass. Pathology of the surgical specimen confirmed diagnosis of leiomyoma, a rare scrotal mass.

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Introduction

Leiomyomas are benign mesenchymal tumors originating from smooth muscle cells. Leiomyomas of the uterus were first described in 1854 by Virchow as tuberculum dolorosum [1]. They represent the most common benign tumor of the genital tract in women of reproductive age. Leiomyomas also rarely develop in other locations, such as the scrotum, ovaries,

bladder, lung, vascular structures, and spermatic cord [2]. Extruterine leiomyomas may be classified according to the site of origin as a piloleiomyoma (derived from arrector pili muscles of hair follicles), an angioleiomyoma (from vessel wall smooth muscle), or a genital leiomyoma (derived from tunica dartos of scrotum and myoepithelial cells of the nipple) [3]. The most common subtype of extruterine leiomyoma is the piloleiomyoma, and the least common is the genital leiomyoma [4].

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The first case of scrotal leiomyoma was described in 1858 by Forsters [5]. Scrotal leiomyoma is categorized as a genital leiomyoma; the exact cause of its development is unknown [6]. Scrotal leiomyoma is a rare form of leiomyoma that can arise from the epididymis, spermatic cord, tunica albuginea, or scrotal wall [4]. The tumor is always solitary, and grows slowly over time if not excised. Very few cases are reported in the literature. In a review article by Siegal and Gaffey, they describe only 11 cases in a review of 11,000 cases of scrotal tumors [7].

Case report

A 37-year-old man with a history of left inguinal hernia repair with mesh was referred to the urology service at our institution for evaluation of painless scrotal mass. He initially palpated the mass within his right hemiscrotum 1 year prior, and reported noticeable but minimal growth in the interim. The scrotal mass started to interfere with intercourse, prompting him to seek medical attention. He denied hematuria, dysuria, flank

pain, urinary frequency, hesitancy, incontinence, or urgency. There was no history of additional surgeries or prior trauma. The patient reported no personal or family history of genitourinary malignancy. Physical examination revealed a mobile, firm, nontender, painless mass of approximately 5-6 cm in diameter within the inferior right scrotal sack. Testes on both sides were normal on palpation, without discrete mass or tenderness. There was no inguinal lymphadenopathy.

Imaging and diagnosis

Routine laboratory testing was performed, including complete blood count with differential, blood chemistry, serum α -fetoprotein, and β -human chorionic gonadotropin levels, all of which were within normal limits. Ultrasound of the scrotum revealed a 5.2-cm hypoechoic vascular mass within the right scrotal sack, corresponding to the palpable abnormality (Fig. 1A-C). The mass was circumscribed, oval in shape, and seen inferior to, and clearly separate from the adjacent right testicle,

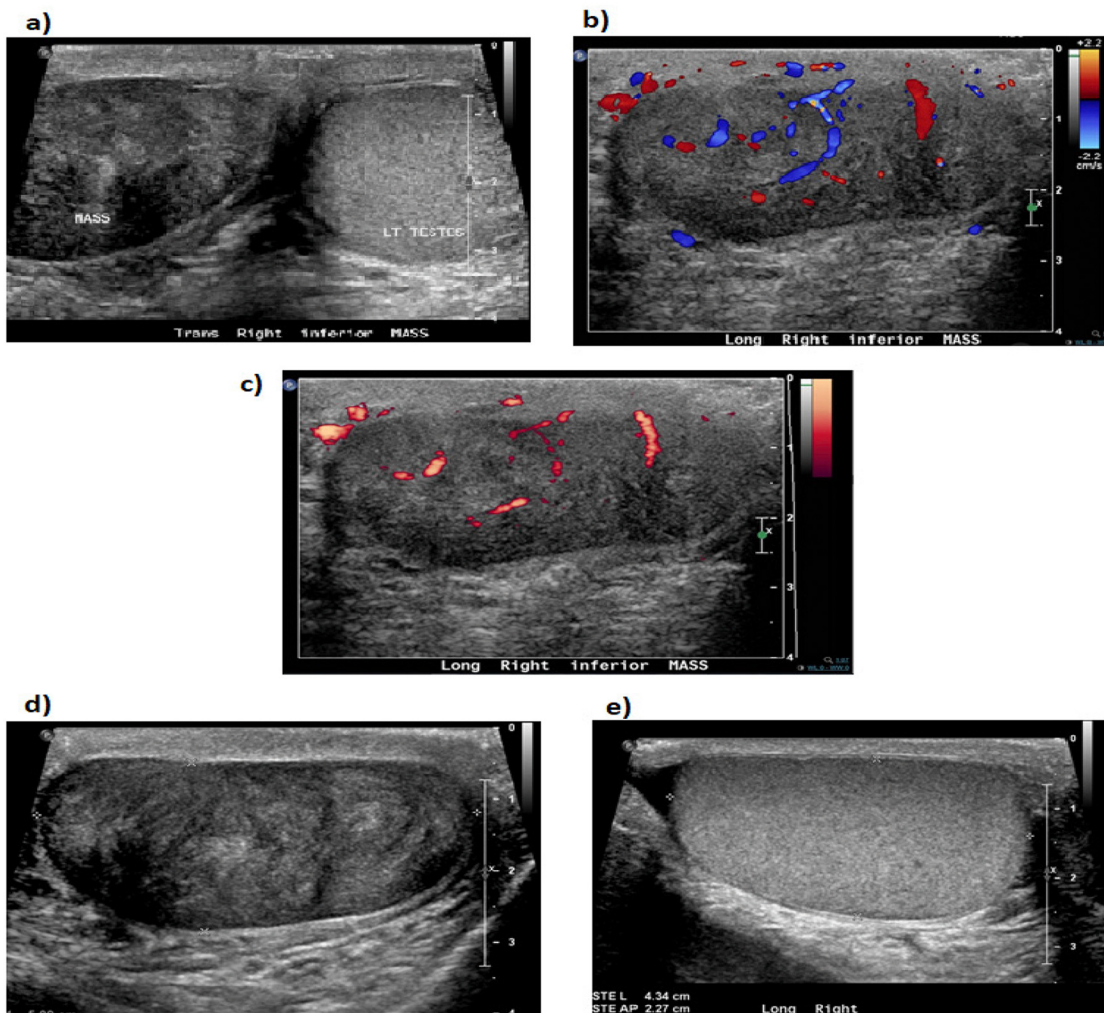


Fig. 1 – (A) Solid hypoechoic scrotal mass corresponds to the area of palpable concern by the patient and is different in echotexture when compared with the left testis in the same image. This mass was shown to be separate and inferior to the right testis (see panel E). (B) Color and (C) power Doppler ultrasound images show moderate vascularity within the mass. (D) Solid heterogeneous circumscribed oval mass in the right hemiscrotum is contrasted with (E) homogeneous echogenicity of the right testis.

epididymis, and other scrotal structures (Fig. 1D-E). In contrast to the normal testicle, the mass was mildly heterogeneous in echogenicity and contained several internal echogenic foci, suggestive of calcifications. No necrosis was observed within the mass.

Based on the broad differential diagnosis, and the continuing discomfort on the part of the patient, surgical excision was planned for pathologic diagnosis and symptomatic relief. A right inguinal exploration and excision of the scrotal mass were performed. A large right scrotal mass was located within the tunica next to the right testis and was found to be adherent to the wall of the right hemiscrotum, with no major vascular involvement. The mass was completely excised and sent to pathology for frozen section evaluation, which revealed no evidence of malignancy. At the same time, the right and left vas deferens were also removed based on patient request. The patient's postoperative course was uneventful.

The pathology report stated that the gross examination of the right and left vas deferens was unremarkable. The right scrotal mass measured $5.5 \times 3 \times 2$ cm and weighed 37 g. Serial sectioning revealed an encapsulated, tan, whorled, rubbery tissue. Histology sections of the scrotal mass revealed a circumscribed, spindle cell neoplasm with mild to moderate cytologic atypia and abundant eosinophilic cytoplasm. Immunohistochemical stains revealed positivity for vimentin, desmin, and smooth muscle actin. The tumor was negative for pan-cytokeratin, CD34, S-100, HMB45, and Melan-A. A Ki-67 immunostain showed proliferation index of approximately 3%. These immunohistochemical findings are consistent with leiomyoma.

Discussion

Scrotal masses are primarily classified as intratesticular or extratesticular, and either solid or cystic. Solid intratesticular masses are malignant in 90%-95% of cases. These include germ cell and non-germ cell tumors, metastatic lesions, lymphoma, and leiomyosarcoma [8]. Extratesticular masses are usually benign, and include lipoma, adenomatoid tumor, papillary cystadenoma, leiomyoma, fibrous pseudotumor, sarcoid granuloma, and polyorchidism. Solid extratesticular masses are malignant in only 3% of cases, consisting of sarcomas, such as liposarcoma, leiomyosarcoma, rhabdomyosarcoma, lymphoma, and metastasis [9]. Of note, testicular cancer is the most common solid cancer in men between 15 and 35 years of age.

Most extratesticular scrotal masses are benign, and majority of these are cystic in composition. Most common of the extratesticular solid masses is a lipoma, which presents as a circumscribed echogenic avascular mass in the scrotum separate from the testis. Magnetic resonance imaging (MRI) is particularly helpful in detecting intralesional fat, which aids in diagnosis of lipomas. Adenomatoid tumor is the most common epididymal tumor and accounts for approximately 30% of all paratesticular neoplasms, second only to lipoma [9]. Papillary cystadenomas have a strong association with von Hippel-Lindau disease, presenting as solid masses with cystic spaces, and are frequently bilateral. Fibrous pseudotumor is a benign proliferation of tissue usually in response to prior

trauma or infection, which may present as a painless solid scrotal mass; the overall appearance is nonspecific and surgical resection is performed for diagnosis [10]. Polyorchidism is a rare condition, presenting with more than 2 testes, most commonly 3 total. All of the testes have the same ultrasound echogenicity, making the diagnosis fairly straightforward. Scrotal involvement in sarcoidosis is uncommon, usually presents with bilateral involvement, and most frequently affects the epididymis with progressively mass-like enlargement.

The majority of malignant tumors in the nonepididymal extratesticular soft tissues are sarcomas that arise from the spermatic cord. The most common scrotal sarcomas are rhabdomyosarcoma (usually presenting in children), liposarcoma, leiomyosarcoma, and malignant fibrous histiocytoma [11]. These are rare neoplasms that present as slow-growing scrotal masses with nonspecific imaging appearance. Intralesional fat is helpful in suggesting diagnosis of liposarcoma. Surgical excision is performed for diagnosis and curative intent. Scrotal involvement with lymphoma may present as unilateral or bilateral testicular masses. Scrotal metastases are usually multiple and tend to present in the setting of widespread metastatic involvement.

Scrotal leiomyomas are some of the least common tumors of the male reproductive tract [5,12]. Typically, scrotal leiomyomas are slow growing, and present in the fifth decade of life [8]. Because of the painless and indolent nature of the mass, patients tend to present 7-8 years after onset of growth [9]. In our case, the patient presented within 1 year of recognizing the mass, which was slowly growing. Physical examination is helpful for assessing scrotal masses, but distinguishing between extratesticular and intratesticular may be difficult. Scrotal ultrasonography is essential in confirming the testicular vs extratesticular origin. Serum markers such as α -fetoprotein, β -human chorionic gonadotropin, and lactate dehydrogenase levels may serve as a useful adjunct.

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. It also allows for detection of any associated features such as calcifications or necrosis. Observing the echogenicity of the lesion on ultrasound assists the physician in evaluating relevant characteristics of scrotal masses, namely, if they are cystic, solid, or both. Scrotal ultrasonography is a reliable method to distinguish between intratesticular and extratesticular scrotal masses, with a 92%-98% sensitivity and 95%-99.8% specificity for testicular malignancy [13,14]. On sonography, normal testes have a homogenous and granular echotexture. The scrotal mass in this case displayed internal heterogeneity and was associated with small calcifications.

In our case, only ultrasound imaging was acquired due to location of the mass in the scrotal sack. MRI, however, is often the most specific imaging for leiomyoma. According to a study done by Dudiak et al, it was found that MRI is the more sensitive and accurate imaging modality in detection and localization of leiomyomas than ultrasound [15,16]. Because of the rarity of this diagnosis in this location, no specific evidence has been presented to compare the utility of the 2 modalities specifically in genital leiomyoma. A case described by Lu et al, however, reports magnetic resonance characteristics in a scrotal leiomyoma that mimic typical

characteristics of leiomyomas elsewhere in the genitourinary tract [17]. Namely, the lesion demonstrates isointense signal on T1-weighted imaging, low signal on T2-weighted imaging, and decreased enhancement relative to adjacent organs, in this case the testes. Definitive diagnosis of leiomyoma requires histologic examination of a resected specimen, looking for features of smooth muscle differentiation. Determining a benign diagnosis is important as it guides the surgical management. Specifically, in this case, and as may be the case in many similar cases, frozen section evaluation and complete excision of the tumor allowed preservation of the testis.

Conclusion

Leiomyoma of the scrotum is a rare benign mesenchymal neoplasm, most often presenting as a painless mass in middle-aged men. This case depicts the typical clinical and radiological features of scrotal leiomyoma. The imaging findings on ultrasound are illustrative of the difficulty in distinguishing leiomyoma from other benign and malignant lesions.

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