Case Report

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Localized giant solitary fibrous tumor of the scrotum: a rare case report and literature review

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Abstract

Background: A solitary fibrous tumor (SFT) is a fibroblastic mesenchymal tumor initially thought to originate from the pleura but that may arise at almost any anatomic site. It is mostly benign, and surgical resection is usually the best treatment option. An SFT involving the scrotum is extremely rare.

Case presentation: We herein report an uncommon case of a 22-year-old man who presented with a huge asymptomatic scrotal mass that had begun growing 3 years before presentation. Contrast-enhanced computed tomography revealed a heterogeneous, well-circumscribed scrotal mass with soft tissue density. No invasion of the surrounding organs, distal metastasis, or lymph node swelling was present. Complete resection of the mass was successfully performed. The specimen was a $14.5 \times 12.0 \times 9.5$ cm encapsulated tumor that weighed 970 g. After pathological analysis, we confirmed the diagnosis of SFT. This diagnosis was based on clinical findings, histological morphology, and immunohistochemistry. No recurrence or metastasis was observed during a 3-year follow-up.

Conclusion: SFTs have an unpredictable clinical course, and they are difficult to diagnose and easy to misdiagnose. A scrotal location is extremely rare. Complete resection of the mass is the treatment of choice and is associated with a high success rate and low recurrence rate.

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Keywords
Solitary fibrous tumor, scrotal mass, conservative surgery, computed tomography scan, immunohistochemical analysis, histology

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Background

The first mention of a solitary fibrous tumor (SFT) was probably by Wagner in 1870,[1] who described an SFT of the pleura. However, it was only later in 1931 that an SFT was described in more detail by Klemperer and Rabin,[2] who reported the pathological characteristics of SFTs.[3] Subsequent studies further investigated the source of SFTs and determined that they probably arise from the mesenchymal cells. Thus, SFTs are most common in the lung pleura, but they can also arise from any other mesenchymal cells of the body located in anatomic sites such as the meninges, nose, oral cavity, pharynx, epiglottis, salivary gland, thyroid, breast, kidney, bladder, and spinal cord.

Because few cases of SFTs have been reported and the clinical course varies among individual patients, there is no consensus on the treatment of SFTs. Possible treatment modalities range from chemotherapy and radiotherapy to simple observation, but most researchers have recommended complete surgical excision with clear margins accompanied by repeat excisions as the best course of action. We herein report a rare case of an SFT of the scrotum, which to our knowledge has been reported very few times in the literature to date.[4]

Case presentation

A 20-year-old man presented with a 3-year history of a painless right inguinoscrotal mass that had been gradually increasing in size for the past 3 years. Physical examination revealed a large, round, tough mass located between the right testicle and the inguinal region, separated from the testis and epididymis. The tumor had smooth surfaces and a limited capacity of mobility. The skin around the mass had a normal appearance and normal temperature but was adhered to the mass. The patient and his parents provided written informed consent prior to any treatment.

A computed tomography (CT) scan revealed a $14.0 \times 13.0 \times 10.0$ cm well-defined, non-uniform inguinoscrotal mass with mainly low density. The mass was moderately enhanced in a non-uniform pattern within which a large amount of tortuous vessels was observed. The testis was normal with a clear boundary to the mass, and no swollen bilateral inguinal or pelvic lymph nodes were observed (Figure 1).

With the patient under general anesthesia, the huge mass was completely removed via a right inguinoscrotal approach with preservation of the testis (Figure 2). The tumor originated from the tunica vaginalis without infiltration to the testis or surrounding tissue. The postoperative pathological examination confirmed that the mass was a mesenchymal tumor with classic characteristics including fibroplasia, mucoid degeneration, small vascular hyalinization, clear boundaries, very few areas of localized tumor necrosis, no nuclear division, and no foci of hemorrhage (Figure 3). Immunohistochemical examination revealed the presence of numerous tumor markers, including CD99+, Bcl-2+, CD34+, and partial PGP9.5+ (Figure 4).

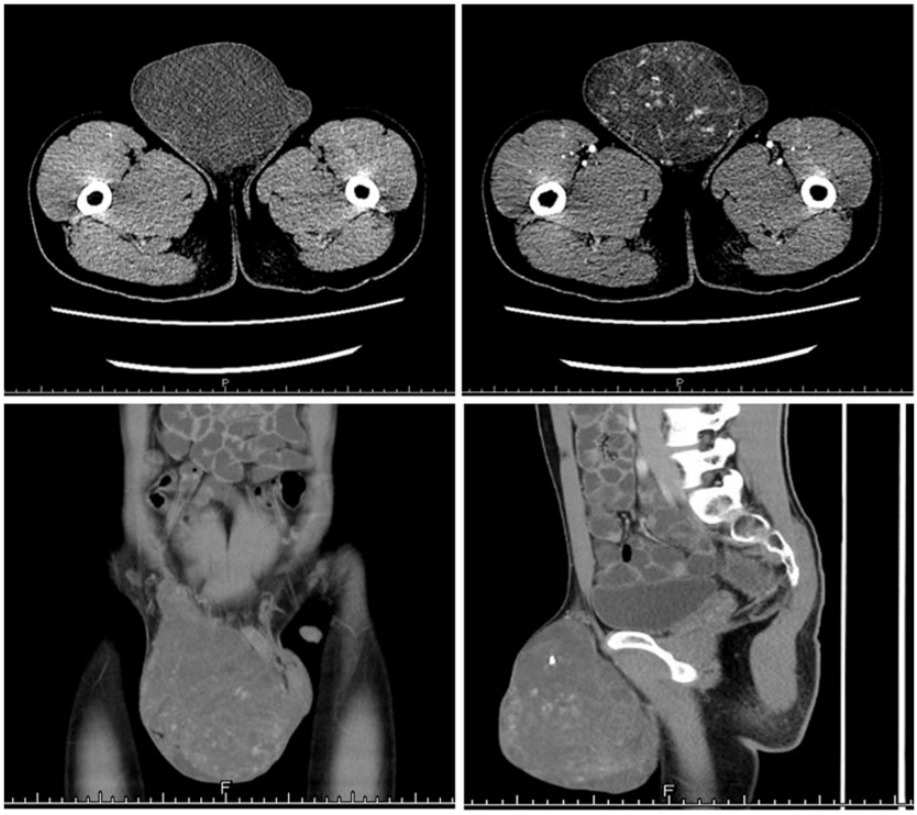

Figure 1. Computed tomography of a 22-year-old man with a giant scrotal mass. Preoperative computed tomography of the abdomen demonstrates a giant (14.5 × 12.0 × 9.5 cm) solid mass in the right side of the scrotum without necrosis. Heterogeneous enhancement is seen during the arterial phase, and large amounts of tortuous vessels were observed.

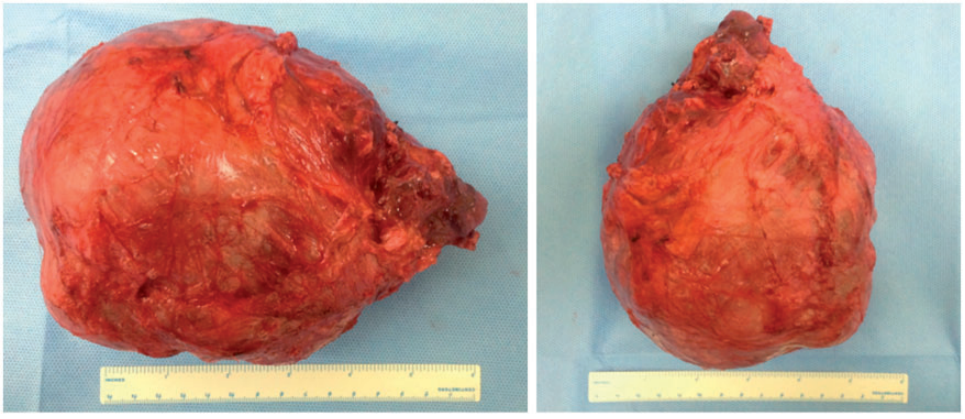

Figure 2. Gross appearance of the tumor during surgery. Large amounts of circuitous blood vessels are extensively distributed on the surface of the mass. The specimen measures 14.5 × 12.0 × 9.5 cm.

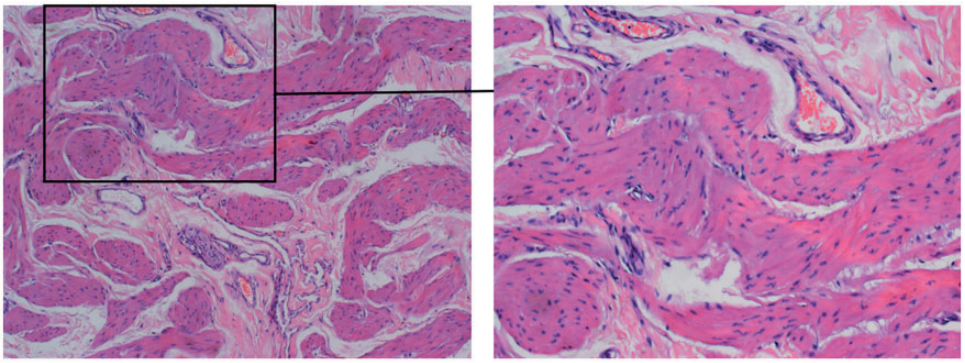

Figure 3. Pathologic examination of the lesion with hematoxylin–eosin staining reveals that the mass is a mesenchymal tumor with classic properties.

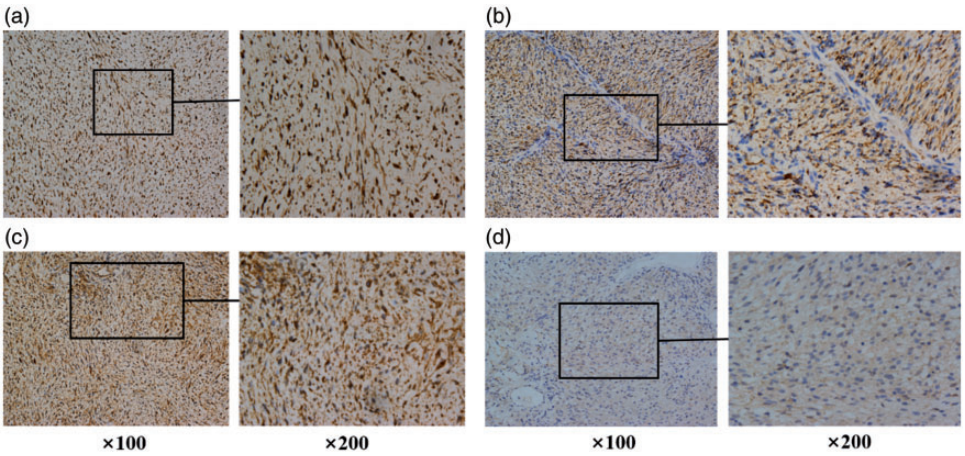

Figure 4. Immunohistochemical examination of the tumor with specific antibodies. Immunohistochemical staining of the tumor section shows (a) strong positivity for Bcl-2 in both the cytoplasm and membrane, (b) strong positivity for CD34 in both the cytoplasm and membrane, (c) strong positivity CD99 expression in both the cytoplasm and membrane, and (d) moderate positivity for PGP9.5 in the cytoplasm and membrane.

The patient remained healthy and developed no local recurrence or metastasis during the 3-year follow-up period. He underwent regular ultrasound examinations each year after surgery.

Discussion

An SFT is a tumor that arises from within the mesenchymal cells of deep soft tissues.[5] Although the first SFT was discovered in the pleural cavity, some statistics now show that 50% to 70% of all SFTs are extrapleural and can occur in the meninges, nose, oral cavity, pharynx, epiglottis, salivary gland, thyroid, breast, and spinal cord; more rarely, they can originate in urogenital organs such as the kidney, bladder, seminal vesicles, and scrotum.[4,6–11] Usually, SFTs located outside the pleura are much

more aggressive.[12] In the present case, however, the SFT in the scrotum showed no local infiltration. Moreover, because SFTs of the scrotum are extremely rare, proper care should be taken to distinguish them from other more commonly occurring masses of the scrotum, such as testicular cancer and inguinal hernias.[13]

Clinical symptoms of urogenital SFTs are mainly determined by the tumor properties: size, location, invasion, and the degree of mucosal integrity. Superficial lesions tend to present as slowly growing painless masses; if any symptoms are present, they are usually caused by the pressure effects on adjacent structures.[14]

Radiologically, an SFT is commonly a large, lobulated, well-delineated, solid, and vascular mass. The mass may exhibit areas of necrosis, hemorrhage, and cystic or myxoid degeneration.[15] In an unenhanced CT image, the mass appears hypodense, with patchy areas probably due to necrosis within the tumor, and mild or marked heterogeneous enhancement that indicates vascularization.[16] Magnetic resonance imaging also plays an important role in the diagnosis of SFTs. The mass is usually isointense or slightly hyperintense in T1 sequences and hyperintense in T2 sequences. Positron emission tomography-CT can indicate a malignancy preoperatively and reveal distant metastases or recurrences postoperatively. Nonetheless, some SFTs have appeared hyperdense on unenhanced CT but with no enhancement under contrast, indicating that diagnosis of an SFT by imaging modalities only is inadvisable and that further pathological examinations are necessary.[8]

Macroscopically, SFTs are usually circumscribed, firm, often partially encapsulated masses measuring 1 to 36 cm in diameter with a mean of 5 to 10 cm.[17] SFTs are usually round or ovoid masses without an obvious pattern surrounded by large amounts of tortuous blood vessels. On cut section, areas of cystic change or hemorrhage may be present. Microscopically, SFTs are characterized by proliferation of uniform elongated spindle cells with various amounts of connective tissue such as collagen and reticular fibers. Smaller tumors tend to be poorly vascularized, while larger tumors show vascularization and infiltration into the vascular system.

Malignant SFTs may have some or all of the following features: infiltrative margins, hypercellularity, nuclear pleomorphism, a mitotic index of >4/10 hpf, and necrosis.[18] Demicco et al.[5] found that patient age, tumor size, and the mitotic index were predictive factors of the time to metastasis and disease-specific mortality.

Immunohistochemical studies have established that tumor markers, including CD99+, Bcl-2+, and CD34+, are generally positive in cases of SFT; thus, they are a helpful modality for the diagnosis of SFTs. Positive staining of CD34+ is indispensable in the diagnosis of an SFT.[19] In the present study, the positive staining of CD99+, Bcl-2+, and CD34+ was demonstrated by immunohistochemistry examinations, confirming the diagnosis of an SFT.

Because of the rarity of SFTs, little is known about their clinical behavior. However, a multicenter retrospective study reported an 81.1% 5-year survival rate and a 30% recurrence rate during a 52-month follow-up; most recurrence was localized.[17] No consensus has been established regarding the treatment plan in cases of recurrence, but different centers have various approaches ranging from chemotherapy with ifosfamide and adriamycin, radiotherapy, and secondary enlarged excision of the recurrent tumor.[20–22]

Conclusion

SFTs have an unpredictable clinical course, and they are difficult to diagnose and easy to misdiagnose. Histological morphology

with immunochemistry is the best way to ascertain a diagnosis of an SFT. Complete radical surgical resection is the treatment method with the best prognosis, and repeat excision upon recurrence is advisable. Even after clean excision, recurrence or metastasis is still possible; hence, long-term follow-up is highly recommended.

Abbreviations

SFT: solitary fibrous tumor, CT: computed tomography.

Availability of data and material

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethics approval and consent to participate

All procedures performed in studies involving human participants were in accordance with the ethical standards of the Ethics Committee of the First Affiliated Hospital of Sun Yat-Sen University (2015-093-03) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from the patient.

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