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Solitary fibrous tumor in the lower limb: A rare case report emphasizing the importance of long-term surveillance

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ABSTRACT

Solitary fibrous tumor (SFT) is a rare mesenchymal neoplasm of fibroblastic differentiation, characterized by unpredictable biological behavior. Although initially described in the pleura, it may occur in extrapleural sites, including soft tissues of the limbs, although this is uncommon. The definitive diagnosis relies on histopathological analysis combined with immunohistochemical markers such as CD34 and STAT6, the latter being highly sensitive and specific for SFT. While SFT often exhibits an indolent course, approximately 10–30 % of cases may develop local recurrences or metastases, reinforcing the importance of prognostic risk stratification and long-term follow-up after surgical resection.

Introduction

Solitary fibrous tumor (SFT) is a rare mesenchymal neoplasm of fibroblastic differentiation, characterized by unpredictable biological behavior. Initially described in the pleura, it can occur in several extrapleural sites, including soft tissues of the limbs, although these presentations are uncommon. Definitive diagnosis relies on histopathological evaluation combined with immunohistochemical markers such as CD34 and STAT6, the latter considered highly sensitive and specific for SFT. Although often indolent, about 10–30 % of cases may exhibit local recurrence or metastasis, reinforcing the need for long-term surveillance.

SFT has an incidence of 0.2–2.8 cases per 100,000 per year, mainly affecting individuals aged 40–70, with no sex preference. It commonly arises in the pleura (30 %), meninges (27 %), and less frequently in extrathoracic soft tissues (2–8 %) [1]. Patients usually present with a slow-growing, painless mass, often in their 50 s [2]. Clinical presentation is nonspecific and varies with tumor location, including symptoms like peripheral edema or pressure-related manifestations such as abdominal pain and urinary symptoms [3]. SFTs in the extremities, especially the thigh, are rare [4,5]. Histologically, SFT consists of spindle cells arranged in a disorganized pattern, with low mitotic activity, mild fibrosis, and prominent vascularization [6]. These tumors often exhibit low mitotic activity and lack significant nuclear pleomorphism and/or necrosis [2]. Diagnosis relies on histopathology and immunohistochemistry, particularly nuclear STAT6 expression [7].

The complex histopathological heterogeneity of these tumors makes radiographic imaging challenging [8]. Imaging characteristics include a well-defined mass that is isodense relative to skeletal muscle, with heterogeneous enhancement on contrast-enhanced computed tomography (CT) [9,10]. On magnetic resonance imaging (MRI), a homogeneous intermediate signal is observed on T1-weighted images [11]. Although most SFTs are benign, approximately 10–30 % of cases may show aggressive behavior, with local recurrences or metastases [12]. The

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definitive diagnosis requires histopathological and immunohistochemical analysis, with positivity for CD34 and STAT6 being important markers for confirmation [13]. Complete surgical resection with clear margins remains the treatment of choice, as it is essential for minimizing the risk of recurrence. Consequently, strict long-term clinical follow-up is crucial to detect potential recurrences or metastases, ensuring optimal disease management [14]. Recent molecular advances have demonstrated that the pathogenesis of SFT is closely related to the NAB2-STAT6 gene fusion, which is considered a molecular hallmark of this tumor [15]. This fusion results in the constitutive activation of the STAT6 pathway, leading to uncontrolled cell proliferation and aberrant differentiation [16]. The detection of this genetic rearrangement through techniques such as RT-PCR and next-generation sequencing has become an important tool in the differential diagnosis of SFT, especially in histologically challenging cases [13]. Furthermore, recent research suggests that the STAT6 pathway may represent a potential therapeutic target for new treatments, opening perspectives for targeted molecular therapy approaches [17].

Clinical reports on SFT are crucial for enhancing our understanding of their presentation, progression, and response to treatment, thereby improving the management of this rare neoplasm. Table 1 presents a compilation of studies describing documented clinical cases in the literature, highlighting the wide anatomical distribution of SFTs, demonstrating its occurrence in different body regions, and reinforcing the need for an accurate and individualized diagnosis.

Case presentation

A 54-year-old female patient from Boa Vista (Roraima, Brazil), under oncological follow-up for ovarian neoplasm, reported pain in the left lower limb following trauma, which progressed with increasing edema. A soft tissue ultrasound revealed a deep intramuscular nodule in the left leg, measuring 70 × 30 × 20 mm.

Subsequent magnetic resonance imaging (MRI) showed an expansile intermuscular lesion in the anterolateral compartment of the middle third of the left leg, well-defined and without invasion of adjacent vascular structures, measuring 8.1 × 3.3 × 2.5 cm (Fig. 1). The initial diagnostic hypothesis was neurofibroma; however, this possibility was ruled out after an incisional biopsy, whose histopathological examination suggested a solitary fibrous tumor (SFT).

The definitive diagnosis was confirmed through immunohistochemical analysis, which revealed a mesenchymal neoplasm composed of spindle-shaped cells arranged in a collagenized stroma, with the presence of hemangiopericytic vessels. The sample exhibited diffuse CD34 expression and focal positivity for STAT6, confirming the diagnosis of SFT (Table 2).

The treatment involved complete surgical excision of the lesion, which was performed on June 9, 2022, the same day as the resection of the ovarian neoplasm. A comprehensive immunohistochemical analysis of the resected specimen confirmed the initial diagnosis of SFT. Postoperative MRI conducted six months after surgery showed no evidence of residual or recurrent disease. The patient continued under strict oncological surveillance, undergoing semiannual imaging studies to monitor for potential recurrence.

Although the initial postoperative course was favorable, a recurrence of the tumor was identified 17 months following surgical intervention, emphasizing the unpredictable clinical behavior of solitary fibrous tumors (SFTs). At present, the patient is alive, remains under regular oncological surveillance, and shows no evidence of disease recurrence. She is of middle socioeconomic status and continues to comply with routine follow-up as part of her ongoing cancer care.

Table 1
Reported cases of solitary fibrous tumors by location.

Location	Case Description	Refs.
Skin	Report of a rare cutaneous SFT, derived from mesenchymal cells, with a challenging differential diagnosis due to its unusual presentation	[18]
Meninges	25-year-old patient with seizures; investigation revealed a meningeal SFT in the left occipital region, treated with complete resection and no recurrence after three years	[19]
Pelvic Region	Asymptomatic 38-year-old woman with a heterogeneous adnexal mass on the right side; resection revealed a primary pelvic SFT confirmed by immunohistochemistry	[20]
Pleura	Patient with a history of blunt thoracic trauma nearly 10 years prior, progressively developing respiratory symptoms; diagnosed with a giant pleural SFT	[21]
Oral Mucosa	Asymptomatic lesion in the oral mucosa resembling a "blister"; histopathological diagnosis confirmed SFT	[22]
Retrorectal Space	First reported case of SFT located in the presacral space, discussing clinical and histopathological diagnostic challenges	[23]
Extremities (Thigh)	46-year-old patient with a slow-growing mass in the left thigh, resected with no signs of recurrence after 12 months of follow-up	[5]
Retroperitoneum	60-year-old male patient with nonspecific abdominal pain; investigation revealed a 14 cm retroperitoneal SFT, treated with complete resection	[24]
Mediastinum	52-year-old patient with chest pain and mild dyspnea; imaging revealed a 9 cm mediastinal SFT, treated surgically	[25]
Orbit	Rare case of orbital SFT in a 35-year-old woman with progressive unilateral proptosis; successful orbital resection	[26]
Nasopharynx	47-year-old man with progressive nasal obstruction; biopsy confirmed nasopharyngeal SFT, treated with endoscopic excision	[27]
Bladder	Report of a bladder SFT diagnosed after obstructive urinary symptoms in a 50-year-old male patient	[28]

Discussion

After the detection of tumor recurrence 17 months post-surgery, the patient underwent a new wide-margin surgical resection, which was successfully performed without complications. To date, the patient remains disease-free under regular follow-up with periodic MRI scans. Given the complete resectability of the recurrent lesion, there was no indication for radiotherapy or systemic therapies.

In addition to the diagnostic challenges inherent to solitary fibrous tumor (SFT), prognostic evaluation is essential for proper clinical management and surveillance. According to the risk stratification model proposed by Demicco et al. [12], the main prognostic factors include patient age, tumor size, mitotic index, and presence of necrosis. Applying this model to the present case, the patient was 54 years old (score 0 for age < 55 years), with a tumor measuring 8.1 cm (score 2 for tumors > 7 cm). Histopathological analysis revealed low mitotic activity (< 4 mitoses per 10 high-power fields, score 0) and absence of necrosis (score 0). The total risk score was therefore 2, classifying the patient as having an intermediate risk of local recurrence and metastasis. This stratification is consistent with the tumor recurrence observed 17 months after surgery, reinforcing the importance of risk assessment to guide follow-up and long-term management in SFT cases.

Although the patient was under oncological follow-up due to a previous history of ovarian cancer, there is currently no established causal association between solitary fibrous tumors (SFT) and epithelial malignancies such as ovarian carcinoma [29]. Reported cases of coexistence are considered incidental findings with no recognized syndromic or genetic correlation [30]. Furthermore, the patient reported a prior traumatic event in the left leg before the tumor was identified. However, there is no evidence in the literature supporting a direct causal relationship between trauma and the development of SFT [31].

Despite limitations in access to molecular testing and advanced diagnostic resources in the region where the case was managed, the diagnosis was achieved through morphological assessment and

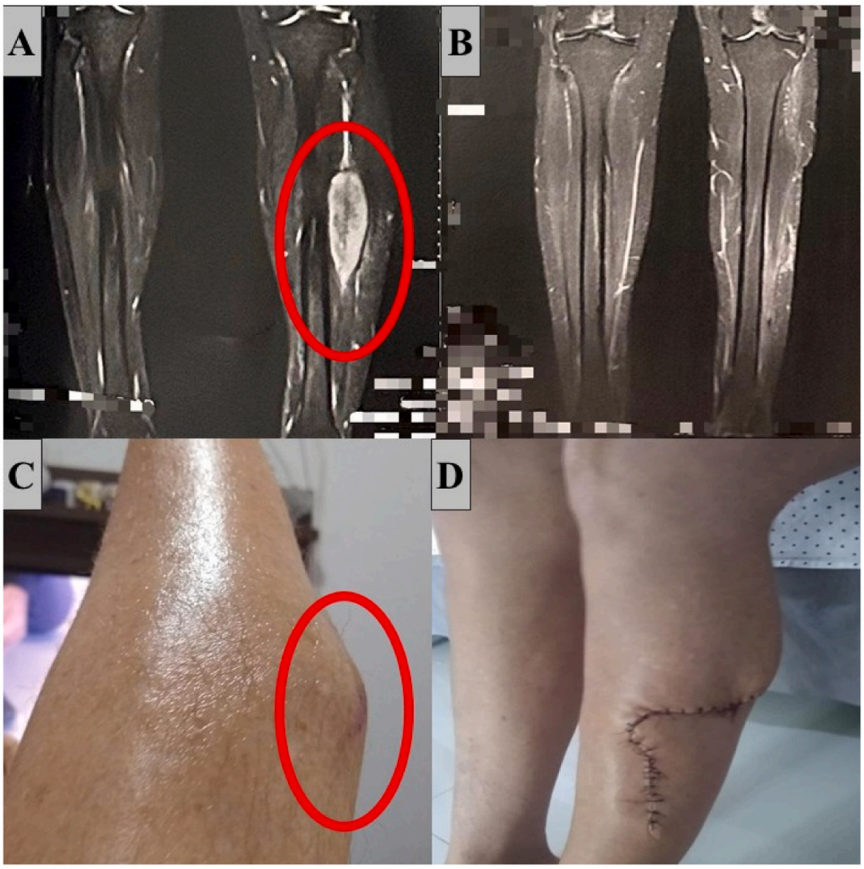

Fig. 1. Magnetic resonance imaging (MRI) and clinical photographs of the patient's lower limb before and after surgical resection of a solitary fibrous tumor (SFT). (A) Preoperative MRI showing a well-defined mass in the anterolateral compartment of the left leg, without invasion of adjacent vascular structures. (B) Postoperative MRI demonstrating the absence of the previously identified lesion, consistent with complete resection. (C) Clinical image of the patient's leg showing the tumor before surgery. (D) Postoperative clinical image displaying the surgical scar after tumor excision.

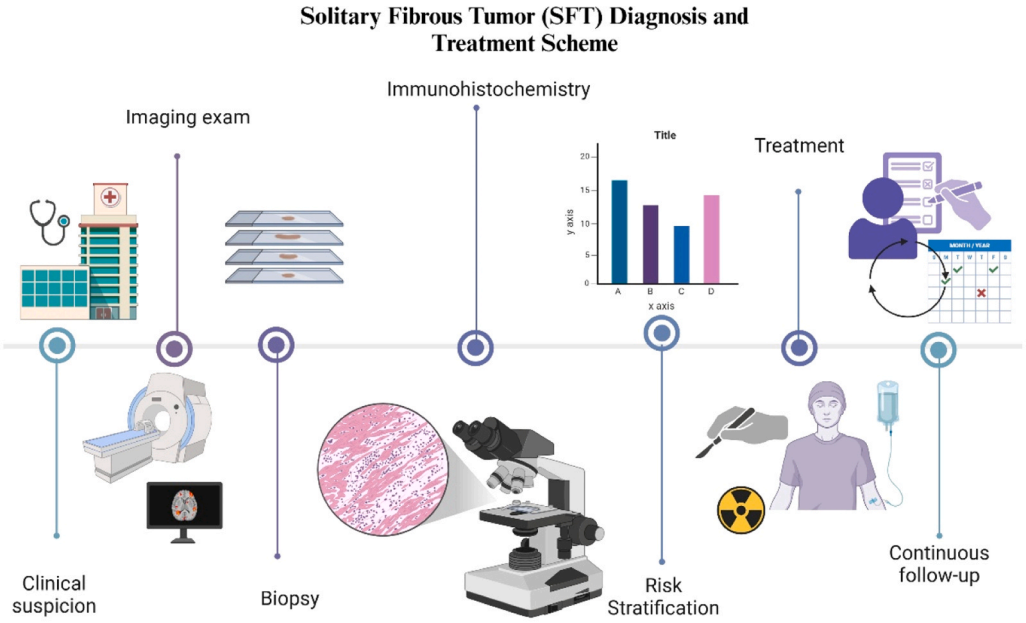

Fig. 2. A timeline illustrating the diagnostic and therapeutic process for Solitary Fibrous Tumor (SFT). Clinical Suspicion: Initial medical evaluation and identification of suggestive symptoms; Imaging Exams: Use of computed tomography (CT) or magnetic resonance imaging (MRI) to detect the lesion; Biopsy: Collection of a tumor sample for histopathological analysis; Immunohistochemistry: Investigation of markers such as CD34 and STAT6 to confirm the diagnosis; Risk Stratification: Assessment of tumor aggressiveness based on histological and molecular characteristics; Treatment: Surgical resection as the main approach, with possible radiotherapy or other therapies in specific cases; Continuous Follow-up: Periodic monitoring to detect recurrences or metastases. Created with *Biorender.com.*

Table 2
Immunohistochemical results before and after tumor removal.

Antigen	Antibody (Clone)	Result Before (Left Tibia)	Result After (Left Tibia)
HHF35	EP35	Negative	Negative
STAT6	EP325	Focal Positive	Focal Positive
S100	EP32	Negative	-
SOX2	EP281	Preserved Expression	Positive
EMA (MUC1)	E29	Negative	Negative
MRO-27	MRO-27	Negative	Negative
AE1/AE3	AE1/AE3&PKC26	Negative	Negative
CD34	QBEnd10	Positive	Positive
DESMIN	DE-R-11	Negative	Negative

The first exam was performed in March 2022 at Laboratórios Santa Maria LTDA - ME, and the second exam was conducted in June 2022 at Laboratório de Anatomia Patológica Nossa Senhora de Fátima LTDA ME.

immunohistochemistry. This underscores the importance of making such diagnostic tools available even in remote areas [32].

Recently, the identification of the NAB2-STAT6 genetic fusion, a molecular marker specific to SFT, has been crucial for diagnosis and differentiation from other mesenchymal tumors [33]. This molecular discovery has been particularly important in histologically challenging cases. Immunohistochemistry with markers such as CD34 and STAT6 allows confirmation of the diagnosis, distinguishing SFT from other spindle cell tumors, such as sarcomas [34].

The diagnosis and therapeutic management of Solitary Fibrous Tumor (SFT) encompass multiple stages, ranging from initial clinical suspicion to post-treatment follow-up. The diagnostic evaluation relies on advanced imaging modalities, biopsy with histopathological analysis, and immunohistochemical techniques, which are crucial for confirming the neoplasm and differentiating it from other mesenchymal lesions. Below is a detailed schematic representation illustrating the key stages of SFT diagnosis and treatment, emphasizing the essential approaches for optimal clinical management.

Complete surgical resection with clear margins is the treatment of choice for SFT, being crucial to prevent recurrences [35]. However, as demonstrated in several clinical cases, such as the patient described, rigorous follow-up is necessary, as the tumor can return after surgery [36]. Early detection of recurrences and metastases through imaging tests, such as magnetic resonance imaging, is essential to ensure treatment success [37]. For patients with aggressive or metastatic tumors, new therapies targeting the STAT6 molecular pathway are being studied, offering new hopes for the treatment of SFT [38].

Although molecular confirmation through NAB2-STAT6 fusion testing was not performed due to the unavailability of this technique in the region, the diagnosis was reliably established based on morphology and immunohistochemical analysis, emphasizing the importance of ensuring access to these essential diagnostic tools even in resource-limited settings.

Conclusion

This report discusses a case of superficial solitary fibrous tumor (SFT), highlighting the challenges associated with the diagnosis and treatment of this rare neoplasm. Despite its low incidence, SFT exhibits significant clinical and histopathological heterogeneity, which can hinder early identification and the establishment of effective therapeutic strategies. Limited access to advanced imaging, molecular testing, and immunohistochemical analysis, particularly in economically disadvantaged regions, represents a major obstacle, leading to diagnostic delays and negatively impacting patient prognosis. Therefore, investments in expanding hospital infrastructure, decentralizing oncological care, and continuously training healthcare professionals are essential measures to optimize disease management. Furthermore, the adoption of standardized post-treatment follow-up protocols is crucial, as SFT may present with local recurrences and metastases even after complete surgical resection. Strategies such as systematic imaging-based monitoring, the implementation of targeted therapies for aggressive cases, and the strengthening of referral and counter-referral networks within the public healthcare system are fundamental to reducing disparities in access to diagnosis and treatment, promoting a more equitable and effective approach. Thus, equity in oncological care should be a priority, ensuring that all patients, regardless of geographic location or socioeconomic status, have access to accurate diagnosis, appropriate treatment, and long-term clinical follow-up. Addressing these disparities requires an integrated effort between the government, healthcare institutions, and the scientific community to improve the prognosis and quality of life of individuals affected by SFT and other rare neoplasms.

CRediT authorship contribution statement

Ana Cássia Silva Oliveira: Writing – review & editing, Investigation. **Pablo Andre Brito de Souza:** Writing – review & editing, Supervision, Investigation. **Bruna Kempfer Bossoli:** Writing – review & editing, Writing – original draft. **Jessica Vanina Ortiz:** Writing – review & editing, Investigation. **Luis Enrique Bermejo Galan:** Writing – review & editing, Investigation. **Felipe Augusto Cerni:** Writing – review & editing, Writing – original draft. **Roberto Carlos Cruz Carbonell:** Writing – review & editing, Investigation. **Manuela Berto Pucca:** Writing – review & editing, Writing – original draft, Supervision, Funding acquisition, Conceptualization. **Geovanna Maria Malachias-Pires:** Writing – review & editing, Writing – original draft, Data curation. **Giovanna Mafra e Silva:** Writing – review & editing, Writing – original draft, Investigation, Data curation, Conceptualization.

Informed Consent Statement

Written informed consent has been obtained from the patient to publish this paper.

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Presentation at a meeting

NA.

Conflicts of Interest

The authors declare no conflicts of interest.

Declaration of Competing Interest

The authors declare that they have **no conflicts of interest** to disclose related to the content of this case report.

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scientific community.

Data availability

Data will be made available on request.

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