Case Report

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Solitary Fibrous Tumor of the Lower Limbs. Presentation of a Case

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ABSTRACT

Introduction: Solitary fibrous tumor (SFT) first described in 1921 by Klemperer and Rabin; it is a rare, primitive mesenchymal cells benign tumor (pleura); called hemangiopericytoma when it is in other regions; difficult to diagnose because of its broad histological spectrum, so it is necessary to resort to immunohistochemical tests. MRI is only useful to establish its location and adjacent lesions; There is no ideal treatment.

Objective: Present a STF case in lower limbs, which by imaging studies was misdiagnosed as a vascular lesion.

Case Report: A 36-year-old female presented with a tumor in her left leg with a 5-month evolution, painful on palpation, without traumatic history, drug addiction, surgical or diagnostic instrumentation or vasculitis. CT angiography was performed.

Result: With the diagnosis of probable Hamartoma versus pseudoaneurysm, Angiology went for surgical intervention resecting the lesion of cystic characteristics, without pulse or phremity nor arterial involvement; evolves satisfactorily, discharging home in three days. Definitive diagnosis of SFT by immunohistochemistry (CD34).

Discussion: Due to the location and contrasted enhancement, it was confused with a vascular lesion; It would have been important to perform a biopsy of the nearby artery and follow-up of the patient, since the size of the lesion according to what has been written in the literature contributes to recurrences; it was not possible to follow up due to the idiosyncrasies of our population and the administrative problems, which made it difficult.

Conclusion: Multidisciplinary management, having the appropriate diagnostic tools and the collaboration of the experienced pathologist are important in uncommon injuries, especially without a history that confirms or suspects their origin.

Keywords: Fibrous Solitary Tumor; Extremities; Immunohistochemistry; Recurrences

Abbreviations: SFT: Solitary Fibrous Tumor

Introduction

Solitary fibrous tumor (SFT) was first described in 1921 an adult pleura, by Klemperer and Rabin; is a tumor originated from primitive mesenchymal cells, located mainly in the chest cavity (pleura); However, it can be found in: nasopharynx, upper respiratory tract, orbit, thyroid gland, kidneys, prostate, spine, peritoneum, liver, intestine and rarely, in the lower limbs [1]. When it is in other regions it has been named Hemangiopericytoma (according to the National Cancer Institute definition); It can usually be confused with a meningioma. Hemangiopericytoma is considered a vascular origin; derives from Zinmerman's pericytes (modified smooth muscle cells, also called mural cells); It is considered a type of cancer that affects blood vessels and soft tissue. Most cases are benign, it is a rare soft tissue tumor, accounting only for 2% of the cases; They rarely affect the musculoskeletal system, with only 22 cases reported in the literature [2]. Its diagnosis is difficult due to its broad histological spectrum, for which immunohistochemical tests (CD-34 and bcl-2) must be used [3-5]. MRI is only useful to establish its location, extension, and involvement of surrounding tissues, as it is not able to distinguish it from other soft tissue tissues [2]. In the extremities it must be differentiated from both benign and malignant lesions, such as: fibrous histiocytoma, fibrosarcoma, neurofibroma, neurofibrosarcomas among others.

Gold found that tumors larger than 10 cm, tend to present metastasis and recurrence, especially when they have a malignant component, that is, an area with marked cellularity and more than 4 mitotic figures without alternation of hypocellular sclerotic areas, so adjuvant treatment with doxorubicin and radiotherapy is necessary, still without establishing the ideal treatment [1,6]. In contrast, tumors smaller than 10 cm, without a malignant component, were successful with surgery alone. Hasegawa, et al [7]. found a range of local recurrence or distant metastasis between 10 to 13% in extrathoracic tumors, with only one case of recurrence in the upper extremity, so it is suggested, a wide resection and long-term follow-up. [1,7,8] Or, post-resection brachytherapy in local recurrences.

Clinical Case

A 36-year-old female with no significant chronic history of degenerative disease; hormonal implant contraception; comes for mass sensation in left leg for 5 months, which has increased in time; painful on palpation (intensity 4/10), with no limitation for walking; denies direct trauma, fractures, drug addiction, surgical or diagnostic instrumentation nor data suggestive of vasculitis. Physical examination revealed a at mid third of the lower limb in external area a 3x3 cm soft, renitent tumor, without murmur or phremitus or changes in temperature or coloration; normal proximal and distal pulses; Normal ROT. The patient was referred to the Oncology department, with normal laboratory studies; tumor markers CA19-9 14, CA 125 6.2, ACE 1, AFP 0.7; immunoglobulins, anticardiolipins, CRP were not requested.

Doppler ultrasound at muscle layers level, there was an ovoid, solid hypoechoic image of well-defined regular contours, with internal vascularity; diameters of 48 x 21 x 21 mm. An angiotomography was requested, evidencing in the anterior compartment, at the junction of the proximal and middle third of the left pelvic extremity a lesion that captures the contrast medium; presents an important enhancement in the arterial phase (234 HU) with little sweep in the venous phase (198 HU); apparently dependent on the anterior tibial artery; no rupture or bleeding data, measuring 59.6 x 24 x20 mm in its largest diameter; It does not condition periosteal or cortical reaction. There was no evidence of dilations or aneurysmal lesions in the abdomen, pelvis, thighs. The diagnosis of probable hamartoma versus pseudoaneurysm is made, so it is evaluated by angiology (Image 1).

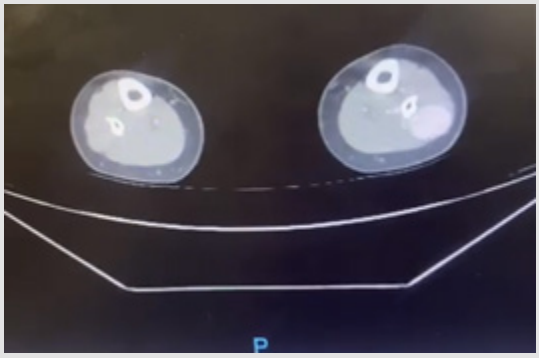

Image 1: CT Angiography of infragenicular vessels; Probable saccular aneurysm versus left anterior tibial artery pseudoaneurysm was suspected.

Surgery was performed, finding a 12 x 8 cm lesion, with cystic characteristics (Image 2), without pulse or phremicus; free anterior tibial artery; the tumor lesion was resected, with arterial and venous integrity at the end of surgery. She evolves satisfactorily so she is discharged to her home three days after the postoperative period. The initial histopathological diagnosis was histiocytoma; The lesion was sent to another pathology unit for immunohistochemical tests where they report: ovoid neoplasm composed of spindle cells that are arranged in fascicles, with indistinct borders; ovoid nucleus and homogeneous chromatin; arborescent vasculature and hyalinized wall. The stroma is collagenized, hyalinized. Immunohistochemistry tests: SOX-10, AML negative. Smooth muscle lactin: SAT-6 (Image 3), CD 99 (Image 4) positive. The positivity of CD34 nuclear expression confirms the diagnosis of solitary fibrous tumor (Image 5).

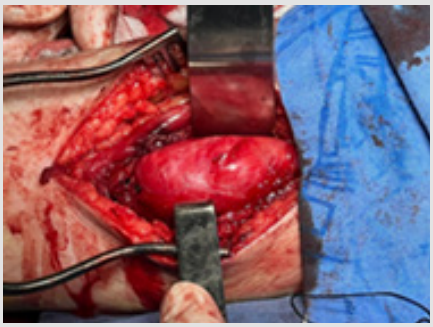

Image 2: Lesion of 12 x 8 cm cystic characteristics.

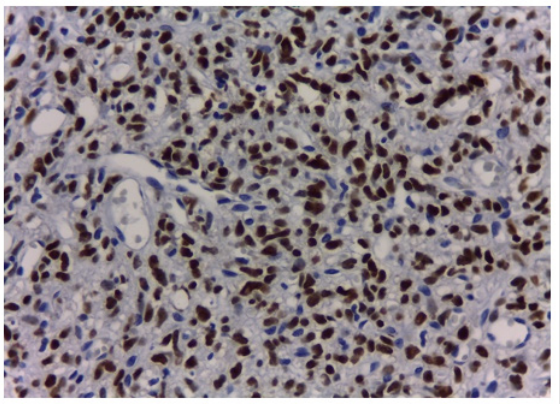

Image 3: SAT 6 Positive nuclear expression.

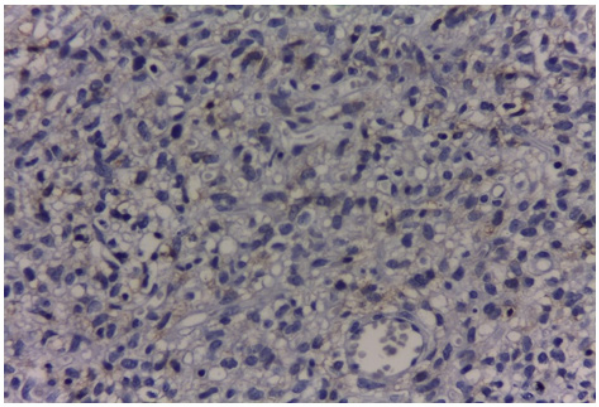

Image 4: CD99 Focal positive.

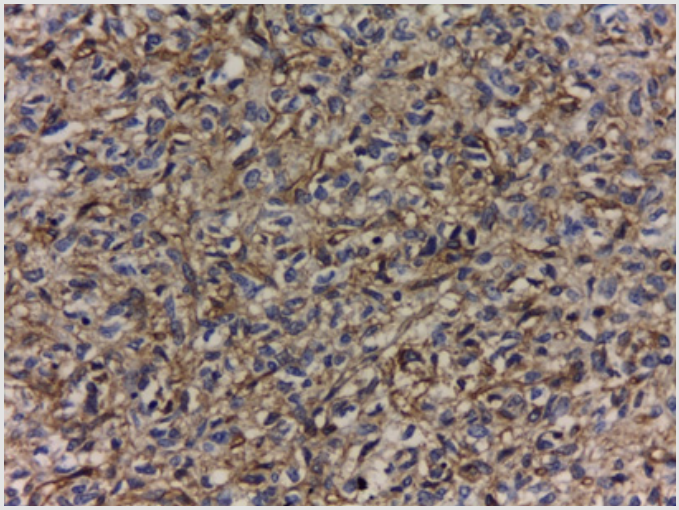

Image 5: CD 34. Positive cell membrane.

Discussion

The SFT, called hemangiopericytoma when located in other sites, it is considered a type of cancer that affects blood vessels and soft tissue. By deriving from the pericytes, it was probably the reason why its location contributed to our patient being diagnosed as an aneurysm or pseudoaneurysm of the anterior tibial artery (even though the patient did not refer to a history of direct trauma, drug addiction, instrumentation in that area or vasculitis data). It would have been pertinent to resect it, take a biopsy of the artery and most importantly, follow up the patient for recurrences, emphasizing the size of the lesion, since as mentioned by Gold et al., when the lesion exceeds the size of 10 cm it tends to recurrency, and according to Hasegawa, in extrathoracic tumors it is up to 13%. In the case of our patient, we cannot verify it due to lack of clinical follow-up, a frequent situation in our environment for cultural and administrative reasons [6,7]. Currently, due to the infrequency of this condition, the ideal treatment has not been established, being important what some authors mention, the adjuvant treatment with doxorubicin and radiotherapy [1,6]; Extensive resection and long-term follow-up, by magnetic resonance imaging (MRI) and/or thoracoabdominal computed tomography, are mandates in the follow up of this tumor [3,8]. That if it is Hamartoma, histiocytoma or SFT or not, the important thing is to establish the definitive diagnosis with all the available tools, because the prognosis and therapeutic behavior is very different in each case, above all, that the patient demands a more specific response.

It is a rare lesion in the extremities, representing only 2% of soft tissue tumors, so we must always keep in mind such clinical entities no matter how rare they may seem. It is currently believed that it has a mesenchymal origin and not mesothelial, although it is more frequent in the pleura and peritoneum. It can originate in any tissue unrelated to serous cavities, being its behavior very different [2,7]. It rarely affects the musculoskeletal system (there are only 22 cases described in the extremities, two in lower limbs); affects men and women equally; It has a broad histological spectrum, which makes its diagnosis more difficult [6]. Immunohistochemical tests are necessary and mandatory. The positivity of CD34 nuclear expression confirms the diagnosis of SFT, as does bcl-2 [3,5]. It is very important to study the wall lesion edges, because being poorly defined or having a peripherical affectation, it is classified as more aggressive. The tumor clinical presentation depends on the size and location; they are usually asymptomatic, however, as in our patient, it caused pain due to the narrowness of the area; or on the contrary, be very symptomatic as those located in the pancreas, which can cause hypoglycemia. A final concept to mention, as they can be developed in any structure, for their discussion it should be referred to as solitary fibrous tumor of the dura, orbita, pleura, spinal cord etc.

Conclusion

Multidisciplinary management, having the appropriate diagnostic tools and collaboration of an experienced pathologist are important in non-common lesions without background, to establish their origin and proper management. We must aise awareness among the patient and family members about the importance of their follow-up, especially in injuries that do not have much experience and their evolution can be catastrophic.

Conflict of Interest

Authors declare no conflict interest.

References

1. Vélez PG, Egea JC, Nogués AC (2008) Solitary adult fibrous tumor in the hip. Rev Esp Cir Ortop Traumatol 52(1): 68-70.
2. Harrington P, Merchant WJ, Walsh ME (1999) Solitary fibrous tumour of the forearm. A rare tumour in an atypical site. J Hand Surg (Br) 24: 370-372.
3. Akisue T, Matsumoto K, Kizaki T, Fujita I, Yamamoto T, et al. (2003) Solitary fibrous tumor in the extremity: case report and review of the literature. Clin Orthop Relat Res 411: 236-244.
4. Hanau Ch, Miettinen M (1995) Solitary Fibrous Tumor: Histological and Immunohistochemical spectrum of benign and malignant variants presenting at different sites. Human Pathology 26: 440-448.
5. Flint A, Weiss Sh (1995) CD-34 and keratin expression distinguishes Solitary Fibrous Tumor (Fibrous Mesothelioma) of pleura from Desmoplastic Mesothelioma. Human Pathology 26: 428-430.
6. Gold J, Antonescu CR, Hajdu C, Ferrone CR, Hussain M, et al. (2002) Clinicophatologic correlates of solitary fibrous tumors. Cancer 94: 1057-1068.
7. Hasegawa T, Matsuno Y, Shimona T, Hirohashi S, Hirose T, et al. (1998) Frequent expression of bcl-2 protein in solitary fibrous tumors. Jpn J Clin Oncol 28: 86-91.
8. O'Connell JX, Logan PM, Beauchamp CP (1995) Solitary Fibrous Tumor of the periosteum. Human Pathology 26: 460-462.

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