VIA MEDICA

CASE REPORT

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Angiomatoid fibrous histiocytoma of the spermatic cord — case report and literature review

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ABSTRACT

In this article, we present a case of a 77-year-old patient with a tumor of the spermatic cord. The tumor infiltrated the structures of the spermatic cord, had a hard and solid structure, and was located in the middle part of the spermatic cord. Since it was found during a procedure, the tumor, along with the spermatic cord and the left testicle, was resected. The entire specimen was sent for histopathological examination. The histopathological examination revealed that the tumor was a non-epithelial neoplasm with a sarcomatous component. Immunohistochemical studies indicated that the tumor corresponded most closely to angiomatoid fibrous histiocytoma. We conducted a review of available literature using a database from 1979 (when this type of tumor was first described) to 2024 and did not find any publications describing a case of angiomatoid fibrous histiocytoma located in the spermatic cord. The search phrases used included angiomatoid fibrous histiocytoma, pampiniform plexus, and spermatic cord. Therefore, it appears that this case is the first known instance of angiomatoid fibrous histiocytoma in this location. In this article, we also reviewed the available literature on the diagnosis and treatment of patients with angiomatoid fibrous histiocytoma and discussed the current approach to diagnosing and treating this rare neoplasm based on our secondary research.

Keywords: spermatic cord tumor, spermatic cord, orchiectomy, sarcoma, angiomatoid fibrous histiocytoma

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Introduction

Malignant tumors of the spermatic cord are rare. Although also uncommon, benign lesions (most commonly lipomas) are usually observed in the spermatic cord. Malignant tumors in this location include myxoid liposarcoma, pleomorphic liposarcoma, well-differentiated liposarcoma, and dedifferentiated liposarcoma, with leiomyosarcoma or rhabdomyosarcoma appearing even less frequently. Malignant tumors of the spermatic cord may also originate from another primary location, such as metastasis from a renal adenocarcinoma [1–3]. However, benign tumors are most commonly found in the spermatic cord and are often mistaken for inguinal hernia. According to the literature, lipomas account for up to 70% of spermatic cord tumors [4, 5].

Case report

A 77-year-old male patient noted a progressively enlarging mass in the left inguinal region during self-examination over the past three months. The patient did not report any pain in the affected area and denied occurrence of any symptoms related to the left testicle. On physical examination, a firm mass was palpable in the projection of the left spermatic cord. Laboratory tests showed that tumor markers (beta-HCG, LDH) were within normal limits.

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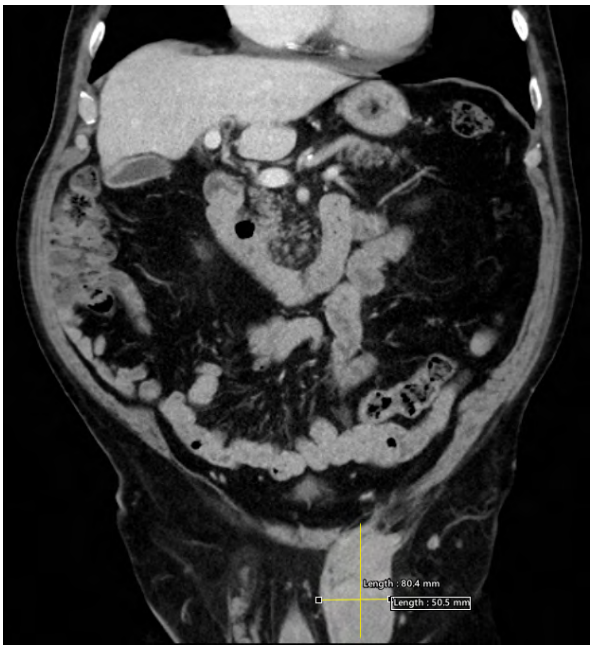

Figure 1. Computed tomography (CT) scan of the abdomen with contrast, venous phase. A tumor of the left spermatic cord, measuring 5 cm in width and at least 8 cm in length, marked with a measurement marker. The lesion was well-demarcated from the surrounding tissues

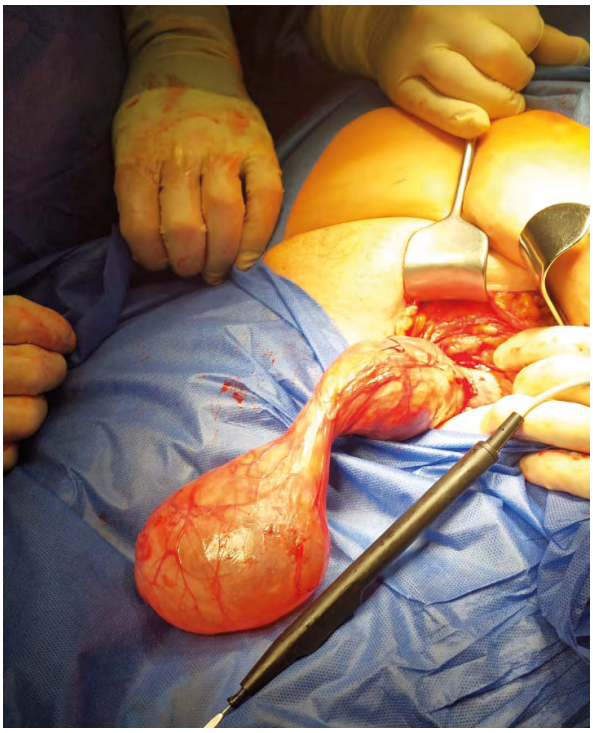

Figure 2. Tumor of the spermatic cord along with the left testicle (testicular hydrocele) — an intraoperative view

An ultrasound study of the inguinal area showed no signs of inguinal hernia, and thrombosis in the femoral or saphenous vein was ruled out. A solid, well-demarcated from the surrounding tissues, hypoechoic mass was found in the left inguinal canal, measuring 57 × 33 × 44 mm. The mass showed minimal vascularization on color Doppler imaging. No pathological inguinal lymph nodes were observed.

For further diagnostic assessment, a contrast-enhanced computed tomography (CT) scan of the abdomen and pelvis was performed. The CT scan identified a solid mass located in the left inguinal region at the scan boundary; it measured 5 cm in diameter and at least 8 cm in length. The mass showed slight enhancement with contrast and was clearly separated from the surrounding tissues (Fig. 1).

The patient was qualified for tumor resection with a left orchiectomy. A longitudinal incision parallel to the inguinal canal was made, cutting through the skin and underlying tissues, along with the extension of the external oblique abdominal muscle. The spermatic cord was dissected. Intraoperatively, a solid, hard tumor of the spermatic cord was found, measuring 8 cm × 4 cm × 3 cm. The tumor was located halfway along the spermatic cord and involved the cord structures. Due to this finding, the entire spermatic cord, along with the tumor and the left testicle, was removed *en bloc*. Additionally, upon releasing the testicle, a hydrocele was identified (Fig. 2 and 3).

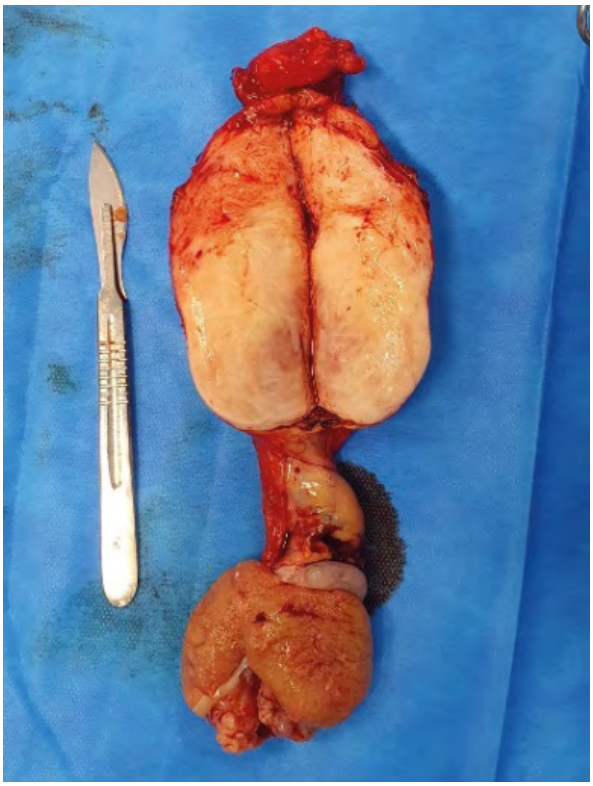

Figure 3. Solid, hard tumor of the spermatic cord along with the testicle — intraoperative specimen. Histopathological examination confirmed angiomatoid fibrous histiocytoma

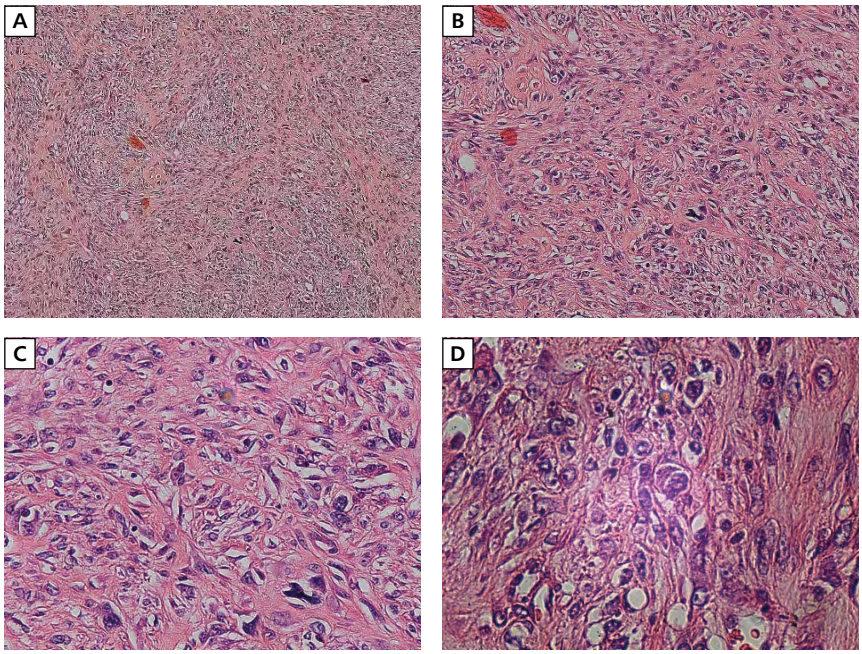

Figure 4. The histopathological image revealed a sarcomatous tissue structure with marked cellular atypia. There was significant nuclear polymorphism, with shapes ranging from spindle-like to nearly round (particularly evident in Figures 4c — 40× magnification and 4d — 60× magnification). The tumor structure itself was fairly uniform. Immunohistochemical analysis showed negative reactions for CD34, SMA, SOX-10, AE1/AE3, AR, beta-catenin, calponin, calretinin, desmin, EMA, inhibin alpha, and S100. A positive reaction was observed for CD99. The Ki-67 proliferation index reached up to 20%, and p53 was approximately 10%*; **A.** Magnification (10×); **B.** Magnification (20×); **C.** Magnification (40×); **D.** Magnification (60×); *The immunohistochemical study was performed at the Department of Pathomorphology, Synevo, Łódź, Poland, by Prof. Radzisław Kordek, MD, PhD

Following removal, the whole specimen was submitted for histopathological examination. The patient was discharged home in good general condition. Histopathological analysis revealed an atypical appearance, most consistent with angiomatoid fibrous histiocytoma (Fig. 4A–D). The histological image showed a tumor composed of spindle cells with polymorphic features, with no visible areas of necrosis. Numerous atypical mitoses were observed. Additionally, a focus on ossification was noted at the periphery of the tumor. Immunohistochemical testing showed the tumor had a positive reaction to the CD99 marker. The proliferative index Ki-67 was approximately 20%, while p53 was around 10%. Other immunohistochemical markers, including CD34, SMA, SOX-10, AE1/AE3, AR, beta-catenin, calponin, calretinin, desmin, EMA, inhibin alpha, and S100, were negative.

A follow-up ultrasound of the scrotum showed the right testicle to be normal. A CT scan of the abdomen and chest revealed no concerning oncological changes. The patient remains under the supervision of a urology outpatient clinic.

Discussion

Angiomatoid fibrous histiocytoma (AFH) is a rare, slow-growing soft tissue tumor, accounting for only 0.3% of all such tumors [6]. It is usually benign but may exhibit local malignancy. The current WHO classification of soft tissue tumors categorizes this tumor as one with undetermined differentiation [7]. The etiology of AFH remains unknown [8]. The tumor is almost always composed of oval and spindle-shaped cells. In one study [9], smooth and striated muscle cells were found alongside cells resembling histiocytes and fibroblasts [8, 10, 11]. Immunohistochemical findings, including the presence of desmin-positive cells within lymphoid proliferations, suggested potential myogenic or myofibroblastic differentiation [6, 8], which led to the introduction of the proposed alternative name “angiomatoid sarcoma” [12].

Due to histological similarities to other tumors, AFH may have previously been underdiagnosed and classified as vascular, fibrohistiocytic, or myofibroblastic tumors [6, 12, 13]. Only after identifying recurrent chromosomal rearrangements resulting in gene fusions

such as EWSR1-CREB1, t(12;22)(q13;q12) EWSR1--ATF1, and t(12;16)(q13;p11) FUS-ATF1, was AFH recognized as a translocation-associated tumor. These translocations are often early events in tumor development, leading to specific chimeric transcription factors that may dysregulate gene expression [14–16]. Among gene fusions, EWSR1-CREB1 is the most commonly observed and found in over 90% of cases [17, 18].

Macroscopically, AFH presents as a hard, multinodular, and hemorrhagic mass with a cut surface ranging from grayish yellow to white. Tumor diameter usually varies from 2 to 4 cm but can reach up to 10 cm (median: 2.5 cm) [19]. In our patient, the tumor measured 10 cm in its largest dimension.

Histologically, the tumor is well-defined, with a lobulated structure surrounded by a fibrous pseudocapsule, which is often incomplete. In approximately 80% of cases, a dense lymphoplasmacytic infiltrate, consisting of lymphocytes and plasma cells, is found around the tumor [6, 19, 20]. In rare cases, atypical morphological features are present, such as cells resembling rhabdomyoblasts or clusters of small cells similar to Ewing sarcoma [21, 22].

Angiomatoid fibrous histiocytoma lacks a specific immunohistochemical profile, which complicates its diagnosis. Approximately half of the cases show desmin expression [6, 10, 23]. CD99 positivity is frequently observed in immunohistochemistry, and other markers such as SMA, EMA, CD68, and CD21 may also be positive [24].

Endothelial markers, such as CD31, CD34, and factor VIII-related antigen, are negative, as are CD35, S100 protein, cytokeratins, and lysozyme [23]. The Ki-67 proliferation index is low, usually around 2–4% [21]. In our patient, the Ki-67 index was up to 20%. Molecular testing, such as fluorescent *in situ* hybridization (FISH) and reverse transcription-polymerase chain reaction (RT-PCR), is essential in the diagnosis of AFH to detect specific fusion transcripts.

This tumor most commonly occurs in individuals under 30 years of age, although cases have been reported in both infants and older adults over 80 years old [11–13]. Angiomatoid fibrous histiocytoma usually appears superficially in the subcutaneous tissue of the extremities, presenting as a slowly growing nodule often mistaken for a hematoma or hemangioma [19].

We reviewed the available literature from 1979 to 2024 and did not find a similar case of this specific tumor located in the spermatic cord. Therefore, our case appears to be the first AFH reported in the spermatic cord. In most patients, the tumor is asymptomatic, though some may report localized tissue swelling in the affected area [25].

In our patient, the initial suspicion was an enlarged lymph node mass. Spermatic cord tumors are often misdiagnosed as inguinal hernias [5]. Other possible locations for this tumor include the trunk, head, neck, and unusual sites such as the brain, lungs, mediastinum, retroperitoneal space, omentum, ovary, vulva, and bones [26–30]. One case of AFH in the spinal cord has also been documented in the literature, involving a 20-year-old male who underwent tumor resection. No distant metastases were detected on MRI at the time of diagnosis. This was the first recorded case of this tumor in the spinal cord, and after 20 months of follow-up, no distant metastases or recurrence were observed [31].

There is no clear association between AFH and other diseases, although cases have been noted in a child with HIV [32] and as a secondary tumor following treatment for other primary malignancies. Examples include a supraclavicular tumor post-treatment for metastatic testicular cancer, as well as cases linked with neuroblastoma and Hodgkin lymphoma [8, 33, 34].

Currently, the recommended diagnostic method for AFH is magnetic resonance imaging (MR) [31]. Differential diagnosis for AFH includes a broad spectrum of tumors, as its histological features may resemble those of Ewing sarcoma, Kaposi sarcoma, or rhabdomyosarcoma [35, 36]. There are reports in the literature that long-term survival and complete remission are possible, even in cases with metastases [18, 20].

The preferred treatment for AFH is wide local excision, as performed in our patient. The tumor should be removed with a wide margin of healthy tissue to prevent local recurrence. Systemic treatment is generally not required for resectable forms [37].

Recently, some authors [38] reported an exceptionally aggressive form of AFH. They presented a case of a 45-year-old man with AFH in the central nervous system (CNS) with disseminated metastases to bones and muscles. This patient was subsequently referred to hospice care.

In cases of metastasis or unresectable tumors, adjuvant radiotherapy or chemotherapy may be considered [39]. Some authors [40], treating a patient with recurrence 18 months post-surgery and lymph node metastasis, administered six cycles of chemotherapy with ifosfamide and adriamycin, followed by three cycles with ifosfamide alone, achieving a favorable response. Another case [38] involved treatment with bevacizumab and temozolomide.

Despite its intermediate malignancy, AFH generally has very good prognosis. Local recurrences occur in approximately 15% of cases [6, 13], and distant metastases are found in fewer than 5% of cases [12, 19, 20], primarily in lymph nodes and rarely in the lungs, liver, or brain [20, 41]. Other authors [19] indicate that the risk of local recurrence is lower with complete excision and ranges from 2% to 10%. In cases of primary soft tissue lesions, there is an 11% risk of local recurrence and a 1% risk of distant metastasis.

Due to the rarity of this tumor, patients should remain under close oncological and/or urological follow-up, as recurrences can occur up to 10 years after the initial diagnosis [42].

Conclusions

1. The described case appears to be the first known instance of this tumor located in the spermatic cord.
2. Currently, immunohistochemical and molecular tests are essential in the differential diagnosis of this tumor, although it does not have a strictly defined immunohistochemical profile.
3. The primary treatment method is surgical resection of the tumor with a wide margin of healthy tissue. In our opinion, in cases of AFH in the spermatic cord, it is advisable to perform a wide excision of the tumor along with the spermatic cord and the affected testicle.
4. Although AFH is considered a tumor of intermediate malignancy, close follow-up is recommended postoperatively to monitor for potential recurrence.

Article Information and Declarations

Ethics statement

As this is a retrospective report of a single case and does not involve experimental procedures, ethical approval from an institutional review board (IRB) or ethics committee was not required. However, all efforts have been made to adhere to the principles of the Declaration of Helsinki and other applicable ethical guidelines regarding patient confidentiality and data protection.

Author contributions

K.K.: manuscript writing, literature review; M.P.: preparation of histopathological documentation; J.G.: provision of histopathological specimens; A.M.: substantive supervision and approval of the final version of the paper.

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Conflict of interest

The authors declare no conflict of interest.

Supplementary material

None.

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