Primary synovial sarcoma of the prostate: A case report and literature review

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Abstract

Synovial sarcoma arising from the prostate is extremely rare. Synovial sarcoma of the prostate is usually identified at a late stage and makes the treatment challenging. Here, we report a case of 30-year-old man with advanced metastatic synovial sarcoma of prostate gland at diagnosis and his poor prognosis.

KEYWORDS prostate, pulmonary metastasis, synovial sarcoma

1 **INTRODUCTION**

Prostatic sarcoma is an uncommon cancer since the prostate has a glandular tissue and the most malignant tumors are adenocarcinomas.^{1,2} Synovial sarcoma (SS) is a malignant mesenchymal tumor of unknown origin that accounts for 8% of all soft-tissue sarcomas.^{3,4} It affects mostly young adults and is more common in the lower extremities; however, it may also arise from other areas, such as the kidney, liver, heart, lung, pleura, esophagus, larynx, middle ear, orofacial or oropharyngeal region, blood vessels, abdominal wall, gastrointestinal tract, and retroperitoneum.⁵⁻⁷ Synovial sarcoma arising from the prostate is extremely rare so far; only a handful of cases (only 8 reported cases) have been reported.⁸⁻¹⁰ Due to its nonspecific presentation, synovial sarcoma of the prostate is usually identified at a late stage.³ This study aimed to discuss the clinical, histological, immunohistochemical, radiographic characteristics and also the treatment of this rare neoplasm. We describe a case of metastatic synovial sarcoma of the prostate.

2 **CASE PRESENTATION**

A 30-year-old man with complaint of urinary obstructive symptoms referred to a Urologist. This symptom with additional progressive suprapubic pain, irritation began from 6 months ago. There was no particular family history of cancer or occupational hazard. On physical examination, there was an irregular large prostate at digital rectal examination. Ultrasound showed an enlarged prostate measuring $48 \times 56 \times 57$ mm and 70 cc volume increase with heterogeneous echogenicity. The urine residue estimated by 20 cc. Magnetic resonance imaging (MRI) revealed malignant appearance of a mass with high signal intensity in T2-weighted MRI, that originate in the prostatic fascia between rectum and bladder and extension to the floor of bladder in addition to anterior sacrum lymphadenopathy with heterogeneous enhancement and necrotic areas within the mass in favor of prostate sarcoma. The patient underwent ultrasound-guided transrectal needle biopsy. Pathologic examination reported Vast multicellular neoplasia with a fascicular pattern of spindle-shaped cells

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with an elongated nucleus and little cytoplasm and some delicate collagen fibers in between, with abundant mitosis and areas of necrosis in the prostate tissue and adjacent to a cross section of the rectal tissue in favor of malignant spindle cell tumor compatible with high-grade synovial sarcoma of the prostate (Figures 1 and 2). In the immunohistochemical examination, TLE1 and EMA were positive and S100, CD34, P63, PSA, and CK7 were negative which confirmed the diagnosis of synovial prostate sarcoma. At metastatic work up, thoracic and abdominopelvic computed tomography (CT) scan and whole-body bone scan were done. There were multiple lung metastasis (Figure 3) and some blastic foci in the left ilium. Also, the prostate was 52×73 mm, larger than normal, which this eccentric prostate enlargement was indicative of prostate tumoral lesions. Whole-body bone scan reported an abnormal uptake in the left iliac crest in favor of bone metastasis (Figure 4). At laboratory study, serum prostate-specific



FIGURE 1 (A) EMA (epithelial membrane antigen) is also focal positive in neoplastic cells, (B) TLE1 (transducing-like enhancer of split1) expression, (C) PSA, prostate-specific antigen, and (D) P63-positive cells



FIGURE 2 Pathologic analysis revealed intertwining fascicles made up of uniform spindle cells with an elongated nucleus and little cytoplasm (high nuclear-cytoplasmic ratio). The mitotic rate was high. Necrosis was seen. (A) spindle cells proliferation hematoxylin and eosin stained (H&E),×4,(B)×10 (C–F)×40

antigen (PSA) was 5.94 ng/ml (normal 4 or less). Other laboratory data were as follow:

Complete blood count, liver function tests, kidney function, and urine analysis tests were normal. Alkaline phosphatase=430 Lactate dehydrogenase=427.The patient presented in our clinic and recommended palliative chemotherapy with MAID regime (Ifosfamide, Doxorubicin, Dacarbazine, and Mesna) every 3 weeks. The patient received 2 cycles of this regimen and refused continue treatment. He died 18 months later.

3 | DISCUSSION

Synovial sarcomas (SS) account for 8% of all soft-tissue sarcomas, and unlike other soft-tissue sarcomas, occurs in young adults (15–40 years old). However, it can occur across a wide age range.^{4,6} SS are relatively common in the distal extremities(about 90%) but they can occur in a variety of unusual locations.⁹SS rarely occurs in the genitourinary system, and if occurs, it is more common in the kidney. Primary prostatic sarcomas are rare tumors (less



FIGURE 3 Chest CT-scan, black arrows show pulmonary metastasis

than 0.1%).¹⁰ SS are frequently small (<5 cm) at presentation; however, most patients such as our patient have a history of obstruction symptoms.⁷Pain is the most common chief complaint rather than an enlarging mass. Our patients also had suprapubic pain for 6 months before diagnosis. According to other reports, prostatic sarcoma occurs in relatively younger adults.^{3,11}To the best of our knowledge, this study presents the youngest prostatic synovial sarcoma.

It is common for a mass of SS to be present for several years before being noticed; therefore, there is a great metastatic potential most likely to the lungs and lymph nodes.^{6,12}

PSA level is often elevated in prostatic adenocarcinoma and in SS of the prostate, due to its non-epithelial origin, serum PSA level may be normal¹³ Our patient had a PSA level of 5.94 ng/ml.

In imaging studies, SS of the prostate usually is a large but inhomogeneous lesion and easily distinguishable from surrounding tissues.¹⁴ Both solid and cystic components of the tumor explain the signal intensity change reported in the literature such as hemorrhage, fluid-fluid levels, and areas of hyperintense, hypointense, and isointense signals relative to fat on T2-weighted MR images.¹⁰In our case, the tumor showed high signal intensity in T2-weighted images. Valenzuela et al showed that most SS were heterogeneous intermediate signal intensity on T1-weighted images, high signal intensity on T2-weighted images, and heterogeneous contrast enhancement.¹⁵

In pathologic evaluations, SS arises from primitive mesenchymal cells which look similar to synovial cells under light microscopy.¹¹ The tumor may have biphasic, monophasic, or poorly differentiated patterns, depending on the proportions of epithelial and spindle cells.⁹ Monophasic is the most common (50%-60%) and consists of only mesenchymal spindle cells. Biphasic is the second common type (20%-30%), which is made of nests of epithelioid cells surrounded by malignant spindle cells.^{6,11} Poorly differentiated SS is the least common subtype (15%-25%).⁶ In immunohistochemical analysis of synovial sarcoma vimentin is positive, epithelial membrane antigen is focally positive and alpha-smooth muscle actin, desmin, and S-100 Protein are negative.⁴ Transducin-like enhancer of split 1 (TLE1) is also an excellent discriminator of synovial sarcoma from other similar sarcomas, such as malignant peripheral nerve sheath tumor.¹⁶



FIGURE 4 Whole-body bone scan showed increased tracer uptake in the left iliac crest (metastasis)

A specific gene translocation, t(X;18) (p11.2q11.2) is found in 90% of patients with synovial sarcomas.¹⁷ This translocation between the SYT gene on chromosome 18 (18q11.2) and one of the three SSX genes on chromosome X (Xp11.2) produces an SYT-SSX fusion transcript that is believed to underlie synovial sarcoma pathogenesis and can be detected by reverse transcriptase-polymerase chain reaction (RT-PCR).^{8,17} On the contrary, the fluorescent in situ hybridization (FISH) method can also be used to identify SYT gene rearrangements.⁸

Due to the rarity of SS of the prostate, treatment protocols have not been clarified. Sarcomas are resistant to radiotherapy, and their response to chemotherapy is poor. Aggressive surgical resection must be considered based on the patient's age and the absence of distant metastases and lymphadenopathy.⁴

In conclusion, diagnosis and treatment of SS of the prostate is challenging due to its nonspecific clinical and radiological data and low incidence. Treatment decisions are made based on local extension, tumor staging, and consideration of risk-benefit analysis and precise clinical and radiological follow-up in terms of recurrence is mandatory.

AUTHOR CONTRIBUTIONS

A.E.T. supervised the case report and contributed to the final version of the manuscript. Z.P.F. contributed to the

interpretation of the results and took the lead in writing the manuscript. R.E. provided the pathologic images and contributed to the interpretation of them. All authors discussed the results and commented on the manuscript.

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Not applicable.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data sets used and/or analyzed during the current study are available from the corresponding authors per request.

ETHICAL APPROVAL

This study was conducted based on the ethical guidelines of the Declaration of Helsinki, 1975, and the patient provided informed written consent for case information and associated images to be published.

CONSENT

An informed written consent form was obtained from the patient.

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