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Case Report

Treatment Process of Primary Prostate Leiomyosarcoma: A Rare Case Report

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Abstract: Prostate sarcoma is an extremely rare malignancy that accounts for only 0.1% of all neoplasms of the prostate gland. Primary prostate leiomyosarcoma (PLSOP) is the most common subtype in adults. Due to the fact that it is an extremely rare malignancy, case reports have been reported frequently and several publications in the form of case series. The number of case reports in the world is less than 200. Our opinion is that publishing such rare diseases and bringing them to the literature will have positive benefits both scientifically and for the patients. We present a patient with PLSOP and discuss the clinical, diagnostic and therapeutic aspects of this rare malignancy.

Keywords: Prostate, Leiomyosarcoma, Cancer, Prognosis

Introduction

Prostate sarcoma arising from the stroma of the prostate gland is an extremely rare malignancy, accounting for less than 0.1% of primary prostate malignancies. Less than 200 cases have been reported in the literature worldwide.1 Primary prostate leiomyosarcoma (PLSOP) is the most common primary prostate sarcoma, constituting 38% to 52% of primary prostate sarcomas in adults.2

Surgery with or without chemotherapy/radiotherapy would appear to be the mainstay of treatment for PLSOP for operable cases, but generally there is no consensus opinion on the best therapeutic approach. Unfortunately most cases of PLSOP at the time of initial presentation tend to be diagnosed in an advanced stage of the disease.

Case Presentation:

A 68-year-old male, with a history of known ischemic cerebrovascular disease, benign prostatic hyperplasia (BPH) was presented to the urology clinic with complaints of difficulty in urinating, dysuria, pollakiuria, and nocturia. Transurethral resection of the prostate (TUR–P) was applied first, due to the history of BPH and normal serum prostate specific antigen (PSA) levels.

The result of TUR–P material pathology was the primary leiomyosarcoma of the prostate. Microscopic examination revealed tumor necrosis, pronounced nuclear atypia and increased mitosis. Tumor cells were immunohistochemically smooth muscle actin and desmin positive, CD34 and CD17 negative.

F–18 fluorodeoxyglucose (FDG) positron emission tomography (PET) imaging was performed. Increased focal FDG uptake was observed in the left side of the prostate gland (SUVmax 8.3). Focal intense increased F–18 FDG uptake was noted in the sigmoid colon (SUVmax 10.1). Pathological F–18 FDG uptake was not observed in other parts of the body. Serum PSA was normal (1.11 µg/L). Colonoscopy was performed due to increased focal FDG uptake in the sigmoid colon. In colonoscopy, a polyp approximately 2 cm in size was detected in the sigmoid colon and a polypectomy was performed. The pathology revealed tumor necrosis, pronounced nuclear atypia and increased mitosis. Tumor cells were immunohistochemically smooth muscle actin and desmin positive, CD34 and CD17 negative.
result of the polypectomy material was reported as tubulovillous adenoma. Since the tumor was considered inoperable it was planned to give 4 cycles (12 weeks) of neoadjuvant epirubicin–ifosfamide chemotherapy regimen to the patient who had no signs of systemic metastasis and ECOG score of 1. Response evaluation with multiparametric prostate MRI was planned after 4 cycles of chemotherapy. After 4 cycles of neoadjuvant chemotherapy, the tumor size was stable. Radical cystoprostatectomy and Bricker diversion were performed because there was no evidence of systemic spread and the tumor size did not decrease to the desired level with chemotherapy. He was also evaluated by radiation oncology after surgery for the need for radiotherapy. Thereupon, follow-up was planned for local recurrence with prostate MRI every 3 months for the first 6 months and then every 6 months after the operation. Our patient, who has now completed the post-op 18th month, has no signs of local/systemic recurrence.

Discussion and Conclusion:
Sarcoma of the prostate gland is a rare neoplasm that originates from mesenchymal cells. There are types of sarcoma, leiomyosarcoma originating from smooth muscles and rhabdomyosarcoma originating from skeletal muscles. Prostate leiomyosarcoma is the most common primary prostate sarcoma in adults however prostate rhabdomyosarcoma is the common primary prostate sarcoma in pediatric patients1,2.

Leiomyosarcoma of prostate gland has been reported in patients whose ages have ranged from 2.5 years to 80 years with a mean age of 61 years. The lack of early specific symptoms in PLSOP cause to be presented with more advanced disease at the time of diagnosing. Since it is not of epithelial origin, its diagnosis may be late, as serum PSA levels will be normal. Leiomyosarcoma of prostate gland usually metastases to the lung, and sometimes to the liver as well 2,3,4.

Leiomyosarcoma of prostate generally presents with hematuria, urine retention and lower urinary tract symptoms including urinary frequency, urinary urgency and poor flow. The symptoms may mimic benign prostatic hypertrophy clinically. Digital rectal examination may reveal an enlarge, firm or hard prostate which may feel benign or has extended to capsule or around tissues. Blood tests and urine may be checked for laboratory investigations. Urinalysis may be normal or have evidence of haematuria and infection but these are not specific to diagnose leiomyosarcoma of prostate. Serum PSA may be normal or raised but it is not a diagnostic tool. Also serum PSA level doesn’t help about progression leiomyosarcoma of prostate.3,5,6 PLSOP is diagnosed by ultrasound guided transrectal needle biopsy or transrectal ultrasound scan of prostate (TRUS) in most patients and less commonly by transperineal biopsy, CT-guided biopsy or suprapubic prostatectomy. It ranges between 2 and 31 cm and is frequently very infiltrative with focal areas of hemorrhage, necrosis and/or cystic degeneration 6,7,8.

Pathologic findings are mostly typical and easy to identify the disease. PLSOP contains some microscopic features that are spindle cells with enlarged hyperchromatic nuclei and increased mitotic activity. Most cases tend to have high grade features on microscopic examination. These includes hypercellular, intersecting bundles of eosinophilic, spindle–shaped cells that have variable degrees of nuclear mitotic
activity as well as nuclear atypia. As immunohistochemically leiomyosarcoma of prostate tend to be positive for vimentin, CD44, smooth muscle actin, calponin desmin and keratin whereas it tends to be negative for S–100, cytokeratin, CD117, PSA and CD34 4–7. On cytogenetic study, leiomyosarcoma of prostate shows clonal chromosomal rearrangement involving chromosomes 2,3,9,11 and 19.9

There are multimodality treatment combinations in the management of leiomyosarcoma of prostate. They include surgery, pre or postoperative radiation therapy, and neoadjuvant or adjuvant chemotherapy. However there is not any standard treatment recommendations. Operable leiomyosarcomas of prostate should be treated with surgery, followed by radiation therapy and/or adjuvant chemotherapy. Bulky leiomyosarcomas of prostate may be operated with neoadjuvant (preoperative) chemotherapy with or without radiotherapy. Curative surgeries include radical retropubic prostatectomy, radical cystoprostatectomy, suprapubic prostatectomy. In patients with inoperable or disseminated disease, systemic chemotherapy may be helpful to induce clinical responses, but remission is rare. There are different chemotherapy regimens including anthracycline (doxorubicin or epirubicin)—based combinations with alkylating agents (cyclophosphamide, ifosfamide, or dacarbazine) and/or vinca alkaloids (vinblastine or vincristine). 2,3,4,10, 11,12,13 Epirubicin, which is less cardiotoxic than doxorubicin, was preferred because the patient had a history of ischemic cerebrovascular disease and an ejection fraction of 50% was detected in echocardiography.

The prognosis of leiomyosarcomas of prostate is poor, because of being aggressive and having high rates of being recurrence or metastasis. Leiomyosarcomas of the prostate gland are aggressive tumours with a median survival of 3 to 4 years and they tend to recur as well as metastasize to the liver and the lungs 16. The cases are with localized disease who have undergone surgical resection with negative margins may have improved prognosis. The prognostic factors of PLSOP is the stage of disease and positive surgical margins after surgery. With the reason of having high risk of death, leiomyosarcoma of prostate may be one of the most aggressive and poorly prognostic malignancies involving the prostate 3,7,14.

Informed consent was obtained from the patient.

Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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