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Table of Contents

Original Articles

Identification of the Physiological Dimension and Self-Concept among Husbands of Iranian Women with Mastectomy; a Directed Content Analysis	06
Marzieh Beigom Bigdeli Shamloo, Nasrin Elahi, Marziyeh Asadi Zaker, Kourosh Zarea, Armin Zareyan	
Tumor–Stroma Ratio in ER+/HER2– Breast Cancer: Is it a Tool for Treatment Decision?	14
Choukri ELMHADI, Mohammed Allaoui, Meryem Zerrik, Mohammed Oukabli, Rachid Tanz, Mohammed Ichou	
Prevalence of BRCA1 and BRCA2 Mutations Among High–risk Bahraini Patients with Breast Cancer	22
Zain Bukamal, Amal AlRayes	
Survival Outcomes of Post–mastectomy Breast Cancer Patients Treated with Hypofractionated Radiation Treatment Compared to Conventional Fractionation –a Retrospective Cohort Study	26
Ciniraj Raveendran, Suma Susan Meloot, I Yadev	
Variants of Human Mucin Genes in Clear Cell Renal Cell Carcinoma and their Potential Prognostic and Predictive Values	35
Jamal Zekri, Mohammed A. Baghdadi, Abdelrazak Meliti, Turki M. Sobahy, Saba Imtiaz	
Study of Efficacy and Toxicity of Capecitabine Maintenance After Response to Docetaxel, Cisplatin, and 5–Fluoracil–Based Chemotherapy in Advanced Carcinoma Stomach	40
Udip Maheshwari, Pankaj Goyal, Varun Goel, Nivedita Patnaik, Venkata Pradeep babu koyyala, Krushna Chaudhari, DC Doval, Vineet Talwar	
EGFR Expression in Gallbladder Carcinoma in North Indian Population	47
Vikash, Vikas Kailashiya, Mohan Kumar, Puneet	
Does the Nightmare of Distressing Complications of Groin Dissection Over with “River Flow” Incision? – Experience of 240 Dissections from Tertiary Referral Oncology Centre, India	53
M D Ray, J R Jeena Josephin, Premanand N	

Review Article

Peptic Ulcer Disease and its Treatments and Risk of Pancreatic Cancer: a Meta–analysis	61
Nasser Alkhashaym, Goot Albuainain, Tuqa A AbuShaheen, Mohammed Y. Alshami, Ali S Almutairi, Ayman Ahmed Sakr, Ayat S Almuhayshi	

Case Reports

Treatment Process of Primary Prostate Leiomyosarcoma: A Rare Case Report	70
Denis Cetin, Mustafa Murat Mdk, Mustafa Mustafayev, Burcak Karaca	
Metastatic Small Cell Carcinoma of a Male Breast: A Case Report and Review of the Literature	74
Nadin Shawar Al Tamimi, Yousra Bennouna, Mohammed El Fadli, Rhizlane Belbaraka	
A Rare Tumor in Adulthood: Extrapneumatic Pancreatoblastoma	79
Ugur Topal, Begm alm Grbz, Hasan Bektas	

Conference Highlights/Scientific Contributions

News Notes	84
Advertisements	86
Scientific events in the GCC and the Arab World for 2023	87



Case Report

Treatment Process of Primary Prostate Leiomyosarcoma: A Rare Case Report

Denis Cetin¹, Mustafa Murat Midik¹, Mustafa Mustafayev¹, Burcak Karaca²

¹Department of Internal Medicine, School of Medicine, Ege University, 35100, Bornova, Izmir, Turkey.

² Division of Medical Oncology, Tulay Aktas Oncology Hospital, School of Medicine, Ege University, 35100, Bornova, Izmir, Turkey.

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 adults²

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

Corresponding Author: Denis Cetin

Address: Department of Internal Medicine, Ege University Hospital, 35100, Bornova, Izmir, Turkey.

E-mail: cetindenis@gmail.com, mmidik@yahoo.com, mustafa.mustafayev@ege.edu.tr, burcak.karaca@ege.edu.tr

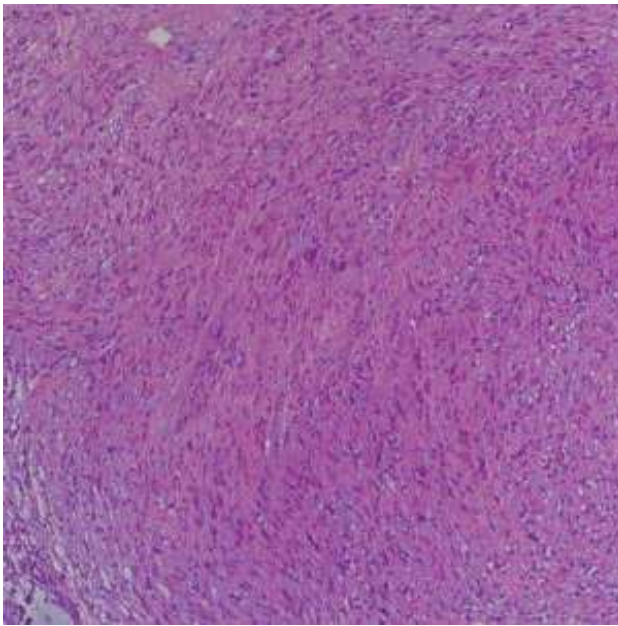


Figure 1: Leiomyosarcoma composed of atypical and hyperchromatic neoplastic spindle cells (Hematoxylin and eosin stain)

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 patients^{1,2}.

Leiomyosarcoma of prostate gland has been reported 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

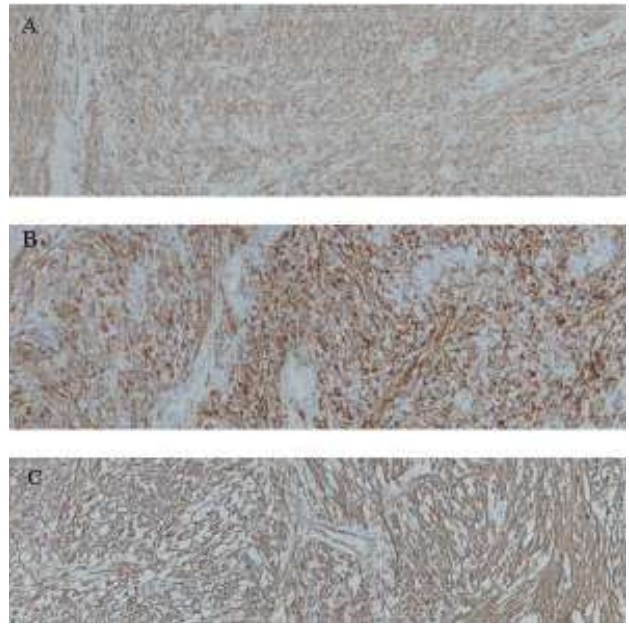


Figure 2: Immunohistochemistry demonstrates that tumour cells express smooth

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 metastasizes 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 CD34⁴⁻⁷. 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 lungs¹⁵. 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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