International Journal of Current Research and Review
DOI: https://doi.org/10.31782/IJCRR.2022.141506

Research Article

IJCRR
Section: Healthcare

Clinical Study of Testicular Leiomyosarcoma: Our Experience

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ABSTRACT

Introduction: Leiomyosarcoma is malignant soft tissue tumor of mesenchymal origin arising from undifferentiated smooth muscle cells. Thera are two types of Leiomyosarcoma of scrotum namely para-testicular and intra-testicular. The Intra testicular tumor is relatively rare. Blood vessels or contractile cells of the seminiferous tubules is believed to be the origin of this rare tumor which is of mesenchymal origin.

Objective: Prospective clinical evaluation of testicular leiomyosarcoma with retroperitoneal mass and metastasis to para-aortic and retro-caval lymph nodes.

Material & Methods: All 21 consecutive patients who were diagnosed as testicular mass on evaluation from August 2018 to July 2021 were included in study after fulfillment of eligibility criteria. Prior to surgery all patients were evaluated. All patients operated with standard method of radical high inguinal orchidectomy, followed by chemo or radio therapy as and when required on further evaluation and histopathology checked. Patients demographic, clinical-pathological, testicular tumor parameters and tumor markers with perioperative data, metastatic parameters recorded prospectively and analysed. Outcome measures were demographic data, tumor response, peri operative data, complications and follow up at 12 months.

Results: In our study out of 21 patients, 13 patients (61.9%) were found to be in stage 1,4 patients (19%) were in stage 2 and another 4 patients (19%) were in stage 3 according to IGCCCG. Only 5 patients (23.8%) were found reteroperitoneal paraaortic and paracaval Lymphadenopathy. Metastasis to liver, lung, brain and major visceral organs were present in 4 of our patients (19.8%).

Conclusion: As less cases have been reported so far, clinical and biological behavior of this tumor difficult to predict. Based on literature review, the treatment of choice for an intratesticular leiomyosarcoma is a radical orchidectomy and clinico-radiological surveillance in cases at stage I. As there are no available data regarding the management of stage II or Stage III disease post radical orchidectomy.

Key Words: Mesenchymal tissue, Leiomyosarcoma, Smooth muscle cells, Testicular tumors, Tumor marker, Reteroperitoneal Lymphdeopathy

INTRODUCTION

Leiomyosarcoma is malignant mesenchymal tumor of soft tissue origin arising from undifferentiated smooth muscle cells. Leiomyosarcoma of scrotum have been classified into para-testicular and intra-testicular, latter being very rare. Primary intra-testicular leiomyosarcoma believed to arise from smooth muscle elements of testis such as blood vessels or contractile cells of seminiferous tubules. So, we are prospectively evaluating clinical study and treatment of testicular leiomyosarcoma with retroperitoneal lymphadenopathy. Our aim of study is to document clinical profile and management of such rare testicular tumors.

MATERIAL & METHODS

All 21 consecutive patients who were diagnosed as testicular mass on evaluation from August 2018 to July 2021 were included in study after fulfillment of eligibility criteria. Prior to surgery all patients were evaluated with CECT abdomen & pelvis for metastasis to para-aortic and retro-caval lymph

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ISSN: 2231-2196 (Print) **ISSN:** 0975-5241 (Online)

Received: 21.06.2022 **Revised:** 10.07.2022 **Accepted:** 22.07.2022 **Published:** 05.08.2022

nodes (Figure1), sonography, chest x ray study and tumor markers {Lactate dehydrogenase (LDH), alpha-fetoprotein (AFP) and beta-human chorionic gonadotrophin(β-HCG)}to prognosticate disease and to know tumor burden. All patients operated with standard method of radical high inguinal orchidectomy (Figure 2), followed by chemo or radio therapy as and when required on further evaluation. Patients were discharged with follow up and level of tumor markers on regular intervals and histopathology checked for Microscopy depicting spindle cells with round or oval-shaped nuclei implicating a storiform whorled pattern with bizarre tumor giant cells and nuclear pleomorphism(Figure 3) and immune histochemistry for smooth muscle actin and vimentin were reviewed(Figure 4), Patients demographic, clinical-pathological, testicular tumor parameters and tumor markers with perioperative data, metastatic parameters recorded prospectively and analysed with categorization and staging according to IGCCCG (international germ cell cancer consensus group). Outcome measures were demographic data, tumor response, peri operative data, complications and follow up at 12 months.

RESULTS

Mean age of male patients in our study was 35 years. In our study out of 21 patients, 13 patients(61.9%) were found to be in stage 1, 4 patients (19%) were in stage 2 and another 4 patients (19%) were in stage 3 according to IGCCCG. 8 patients(38%) were found Tumor size less than 5 cm , another 10 patients(47.6%) were found Tumor size more than 5 cm and rest of 3 patients(14.28%) were found Tumor size more than 10 cm in our study.13 patients(61.9%) were found normal Tumor marker and only 8 patients(38%) were found elevated Tumor marker levels which is unusual in our study.12 patients(57.1%) were found left-sided testicular tumor and 9 patients(42.8%) were found right-sided Tumor in our study. Only 5 patients (23.8%) were found reteroperitoneal paraaortic and paracaval Lymphadenopathy rest of 16 patients (76.1%) were found negative for lymph node in retroperitoneum. Metastasis to liver, lung, brain and major visceral organs were present in 4 of our patients (19.8%) and rest of 19 patients (80.9%) were nonmetastatic. Tumor response was less in large tumor diameter, in presence of necrosis, angiolymphatic invasion, pleomorphism, high mitotic index, atypia. Adjuvant chemoradiotherapy was required in 9 of our patients (42.8%) of which 7 required GC regimen and CYVADIC was required in 2 patients. Rest of 12 patients (57.14%) were not required chemotherapy. Immune histopathological examination confirmed leiomyosarcoma with immune-histochemistry negative for S-100 and myogenic regulatory protein (MyoD1) proteins but was positive for smooth muscle actin and vimentin in all of our patients

DISCUSSION

The exact etiology of testicular leiomyosarcoma is not known. Risk factors include High doses of anabolic steroids, chronic inflammation and prior radiation exposure.[1,2,3] The mean age of the patients is usually above 40 years. The principal presentation is painless testicular enlargement and inguinoscrotal discomfort. The route of spread includes local invasion, lymphatic dissemination and hematogenous metastasis.[4] Most of cases had stage I Tumor. Among few cases four cases had stage III. In all cases that were reported a radical orchidectomy approach was preferred. No adjuvant treatment was given for patients with stage I disease. Adjuvant chemotherapy using gemcitabine plus docetaxel followed by radiotherapy was reported in seven patients who has a high grade tumor and one patient received salvage chemotherapy for distant recurrence.[5] Additional CYVADIC (cyclophosphamide, vincristine, Adriamycin, dacarbazine) chemotherapy was reported in two patients with stage III disease.[6] Five patients underwent radical orchidectomy followed by chemotherapy for a leiomyosarcoma of the testes with the para-aortic lymph nodes metastasis.[7]

CONCLUSION

As less cases have been reported so far, clinical and biological behavior of this tumor difficult to predict. Leiomyosarcoma should be one of the differential diagnosis of seronegative testicular mass. Based on literature review, the treatment of choice for an intratesticular leiomyosarcoma is a radical orchidectomy and clinicoradiological surveillance in cases at stage I. As there are no available data regarding the management of stage II or Stage III disease post radical orchidectomy, hence provision of standard recommendations for therapy is difficult and it should be decided case to case basis.

ACKNOWLEDGEMENT

Sumandeep Vidyapeeth and Dr. Hritik Patel for throughout support for publication and Authors acknowledge the immense help received from the scholars whose articles are cited and included in references of this manuscript. The authors are also grateful to authors / editors / publishers of all those articles, journals and books from where the literature for this article has been reviewed and discussed. I show my humble gratitude towards Sumandeep Vidyapeeth, department of urology team and Dr. Hritik Patel for throughout support in publication

Source of funding

None

Conflict of Interest

author declare no conflict of interest.

Authors' Contribution

Dr. Hritik as laparoscopic surgeon and Dr Anup have worked starting from data compilation to patient operative management tremendously, we have worked as team together with sincerity to fulfill this study work. It was team work and other staff support that made it possible

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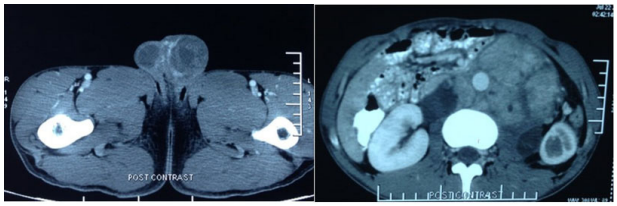

Figure 1: CT KUB: left testicular heterogenous mass & left side retroperitoneal lymphadenopathy.

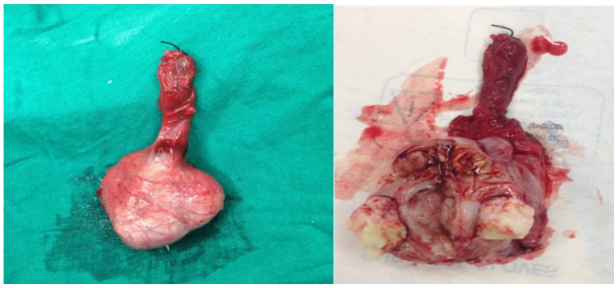

Figure 2: Macroscopically appearance of thetumor, a well-defined encapsulated yellowish white solid mass with hemorrhage and cyst.

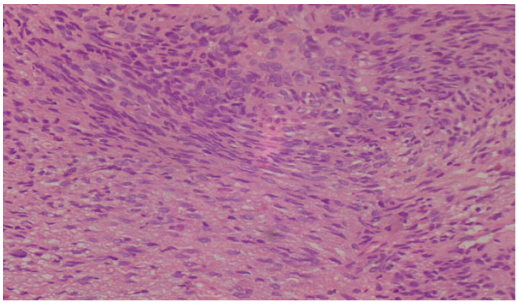

Figure 3: Microscopic picture depicting spindle cells with round or oval-shaped nuclei implicating a storiform whorled pattern with bizarre tumor giant cells and nuclear pleomorphism.

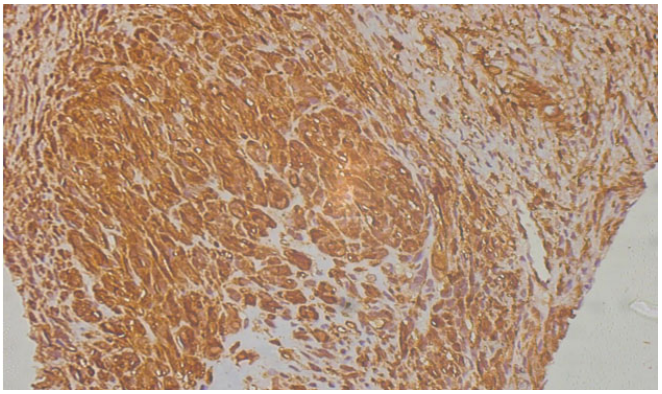

Figure 4: Immunohistochemistry positive for smooth muscle actin and vimentin.