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Dedifferentiated Liposarcoma of the Spermatic Cord: A Case Report

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ABSTRACT

Dedifferentiated liposarcoma of the spermatic cord is an exceptionally rare and aggressive malignancy. Its non-specific clinical presentation and anatomical rarity lead to significant diagnostic and management challenges, with a notable absence of standardized treatment guidelines.

Methodology: This report details the case of a 55-year-old male who presented with a painless left groin swelling. Following initial clinical and imaging assessments, a radical orchidectomy with en bloc tumour excision was performed. Histopathological and immunohistochemical analyses of the resected specimen were crucial for definitive diagnosis.

Key Findings: A 2.5 x 2.5 x 2.2 cm lesion was identified within the spermatic cord. Microscopic examination revealed an infiltrative neoplasm characterized by hypercellular spindle cells within a myxoid background, alongside atypical mitotic figures. Immunohistochemistry showed strong vimentin and S100 positivity, supporting a diagnosis of sarcoma with DDLPS as a strong possibility. The tumour was unifocal with clear surgical margins.

Implications: This case highlights diagnostic pitfalls of spermatic cord DDLPS, often mimicking benign conditions. It underscores the necessity for high clinical suspicion and meticulous histopathological assessment for accurate diagnosis. Radical surgical resection at the earliest with clear margins remains the primary treatment. This report contributes to limited literature, advocating for multi-institutional collaboration and research to establish standardized diagnostic and treatment protocols for this challenging malignancy.

KEYWORDS: Dedifferentiated Liposarcoma, Spermatic Cord, Case Report, Sarcoma, Radical Orchidectomy, Immunohistochemistry, MDM2, Paratesticular Tumour

INTRODUCTION

Dedifferentiated liposarcoma of the spermatic cord is an exceptionally rare and challenging malignancy. Malignant spermatic cord tumours have an annual incidence of only 0.3 cases per million, and liposarcomas account for 3% to 7% of these neoplasms [2]. This rare anatomical location contributes to significant diagnostic and therapeutic complexities [3]. There are fewer than 300 documented cases of spermatic cord liposarcoma in global literature [6].

Clinically, spermatic cord liposarcoma typically presents as a progressively enlarging, painless scrotal or inguinal mass, often observed in the fifth to sixth decades of life [5]. Its non-specific nature frequently leads to misdiagnosis, as it can mimic more common conditions such as inguinal hernias, epididymal cysts, or even lipomas [6]. While imaging modalities like ultrasonography, computed tomography, and magnetic resonance imaging are crucial for initial assessment of mass characteristics, they often lack the specificity to definitively differentiate between benign and malignant lesions or distinguish lipomatous components [2,6]. Primary spermatic cord tumours lack distinctive imaging signs or patterns [6]. This diagnostic challenge underscores the critical need for histopathological assessment to establish a conclusive diagnosis [6].

Accurate histopathological classification is paramount, as the 2020 WHO classification distinguishes between well-differentiated (atypical lipomatous tumour) and dedifferentiated subtypes [6]. Dedifferentiated liposarcoma is particularly aggressive, exhibiting higher recurrence rates, increased metastatic potential, and a significantly lower five-year survival rate of only 28% compared to 85% for well-differentiated liposarcoma [6]. Prognosis is influenced by factors such as tumour grade, anatomical site, and the achievement of clear surgical margins [2,6].

The cornerstone of treatment for spermatic cord liposarcoma is surgical resection. An en bloc resection with radical orchiectomy, aiming for microscopically negative (R0) margins, is the generally accepted and preferred approach [2,3,6]. Due to the rarity of this malignancy, there is a scarcity of level one evidence-based standardized treatment algorithms, with available data primarily derived from case reports and single-institution experiences [2]. Postoperative surveillance with physical examination and cross-sectional imaging is essential, and adjuvant therapies may be considered in high-risk cases, although their overall efficacy is limited and remains an area of study [2,6].

This case report aims to contribute to the limited existing literature by detailing a rare instance of dedifferentiated liposarcoma of the spermatic cord in a 55-year-old male, highlighting the diagnostic complexities and management considerations associated with this challenging malignancy.

PATIENT PRESENTATION

A 55-year-old male, with no known comorbidities, presented to the outpatient department with complaints of a swelling in the lower part of the left groin, first noted two weeks prior to presentation. The patient denied any history of testicular pain or alcoholism.

Upon physical examination, an oval, hard, and painless swelling measuring approximately 3 x2 cm was palpated distal to the external ring of the left inguinal canal. The ipsilateral testis appeared normal, and testicular sensation was preserved. Incidentally, a minimal hydrocele and a Grade 3 varicocele distal to the swelling were also noted during examination. There was no impulse on coughing, and no inguinal lymphadenopathy was identified.

Initial investigations included a computed tomography scan of the abdomen, which incidentally revealed hepatosplenomegaly with portal hypertension. No other mass lesions were detected in the liver, and there was no significant retroperitoneal or paraaortic lymphadenopathy.

SURGICAL INTERVENTION

Following clinical and imaging evaluation, the patient was scheduled for a radical orchidectomy and wide excision of the tumour. The procedure, performed under spinal anesthesia, involved a left inguinal incision to access the inguinal canal. The proximal end of the spermatic cord, which contained a small inguinal bubonocele, was dissected, clamped high, and divided. The spermatic cord, along with its coverings and attached muscles, was meticulously dissected downwards outside the external

spermatic fascia and removed en bloc with the left testis and its coverings. A moderate hydrocele was noted. A neoplasm measuring 3 x2.5cm was identified in the middle portion of the spermatic cord. After tumour excision, plication of the posterior wall was done and the inguinal floor was reinforced with a 15 x 7.6 cm soft polypropylene mesh. Hemostasis was attained and the wound was closed in layers. The excision specimen was sent for histopathological examination.
The post-operative period was uneventful.

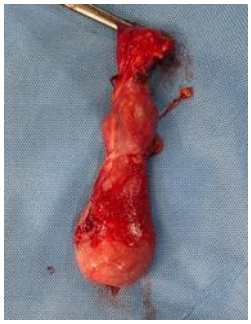

Fig 1: showing gross specimen after radical orchidectomy

HISTOPATHOLOGICAL FINDINGS

Macroscopy:

The radical orchidectomy specimen, including the spermatic cord, measured 13.5 x 4.5x 3 cm. The testis measured 4.2 x 2 x 2.5 cm. A well-circumscribed, capsulated lesion measuring 2.5 x 2.5 x 2.2 cm was observed within the spermatic cord. The cut section of the lesion displayed a solid, glistening appearance with myxoid and hemorrhagic areas. The tumour was situated 4 cm from the resection margin and 2.5 cm from the testis. Both the testis and epididymis were free of neoplastic involvement and appeared viable.

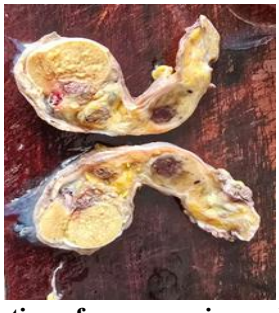

Fig 2: Cut section of gross specimen showing hemorrhagic areas

Microscopy:

Microscopic examination revealed an infiltrative neoplasm encased by a pseudocapsule. The tumour was characterized by hypercellular sheets and fascicles of spindle cells set within a myxoid background, interspersed with hypocellular regions. A nodular architecture, delimited by fibrous septae, was noted in some areas. Elongated, branching, and gaping congested blood vessels were prominent. Individual tumour cells exhibited enlarged, elongated to oval nuclei with irregular nuclear outlines and coarse chromatin. Some cells displayed markedly enlarged and bizarre nuclei, and multinucleated tumour giant cells were present. Atypical mitotic figures were observed, with a mitotic count of 4-5 per 1 mm2. Cytoplasmic vacuolation and clear cytoplasm were noted in some cells. Infarcted areas and stromal hemorrhage were also present. Importantly, there was no evidence of lymphovascular or perineural invasion. Sections from the testis and epididymis were entirely free of neoplasm.

Immunohistochemistry:

Immunohistochemical analysis yielded the following results:

- Vimentin: Diffuse strong cytoplasmic positive.
- CD31, CD34, ERG: Highlighted vascular channels.
- Desmin, SMA, MyoD1, Myogenin, CK, PAX 8, SALL 4, CA9, Calretinin, WT1, CD99, bcl2, STAT6, HMB45, CD45: Negative.
- S100: Highlighted scattered tumour cells.
- Ki67 proliferation index: 15-20%.

Based on the histopathological and immunohistochemical features, a diagnosis of sarcoma was established. De-differentiated liposarcoma was established after MDM2, CDK4 IHC and MDM2 amplification testing. The tumour was unifocal, with a size of 2.5 x 2.5 x 2.2 cm. The resection margin was free of neoplasm, at a distance of 4 cm.

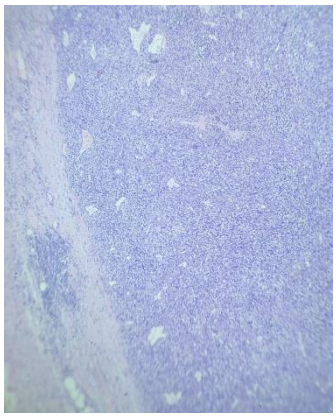

Fig 3 [a]: Hypercellular sheets of neoplastic cells as seen in 4x magnification

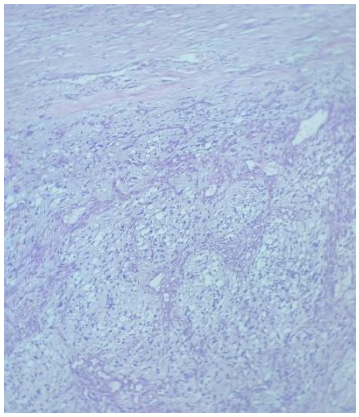

Fig 3 [b]: showing Myxoid areas

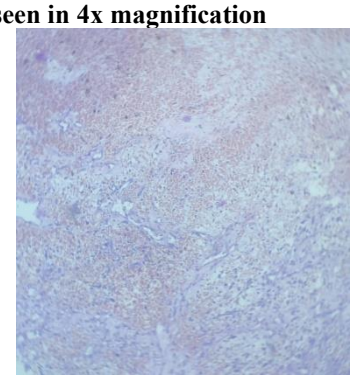

Fig 3 [c]: showing hemorrhagic areas

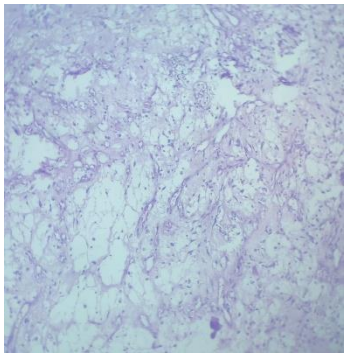

Fig 3 [d]: showing hypercellular areas with vacuolated cells

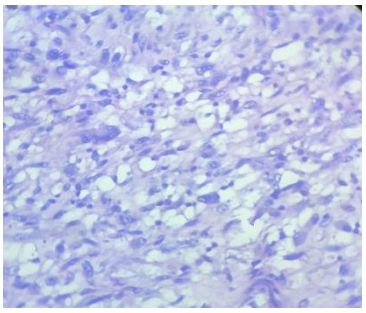

Fig 3 [e]: shows spindle cells with high grade morphology

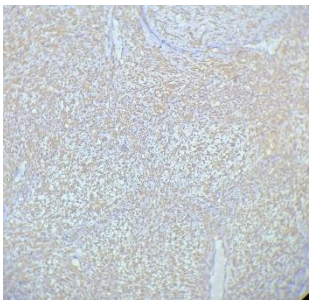

Fig 4 [a]: Diffuse strong vimentin positivity

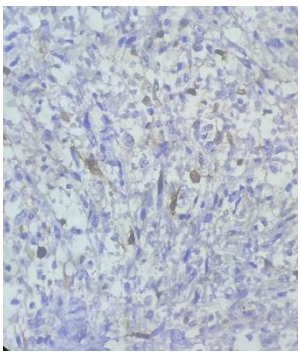

Fig 4 [b]: S 100 – scattered positive cells

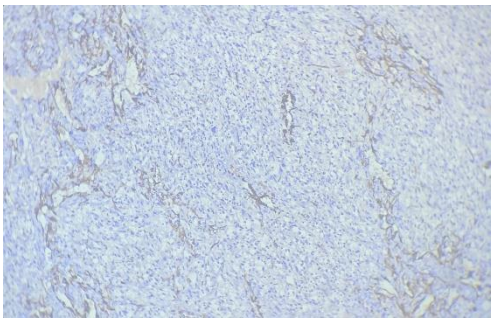

Fig 4 [c]: SMA highlights vascular channels. Negative in tumor

DISCUSSION

Dedifferentiated liposarcoma of the spermatic cord represents a formidable challenge in surgical oncology due to its extreme rarity and aggressive biological behaviour. Our case report of a 55-year-old male with DDLPS of the spermatic cord underscores the diagnostic complexities and highlights critical considerations in

management and future research directions for this uncommon malignancy.

Treatment Modalities and Challenges

The cornerstone of treatment for spermatic cord liposarcoma, particularly the dedifferentiated subtype, is aggressive surgical resection. The standard approach involves en bloc resection with radical orchiectomy and high ligation of the spermatic cord, with the primary objective of achieving microscopically negative (R0) margins [2,6]. The ability to obtain clear surgical margins is a critical prognostic factor, significantly influencing disease-free survival [2,3]. In cases of incomplete resection, reoperation to achieve wider margins has been shown to improve disease-free survival [6]. While testis-sparing surgery may be considered for localized, well-differentiated tumours with clear intraoperative frozen section margins, it is generally not recommended for DDLPS given its aggressive nature [6].

The role of adjuvant therapies, such as radiotherapy and chemotherapy, remains a subject of ongoing debate due to the limited availability of high-level evidence. Their overall efficacy is not definitively established, and no randomized controlled studies exist to guide their use [6]. However, adjuvant treatments may be considered in high-risk scenarios, including cases with positive surgical margins, multiple recurrences, or high-grade tumours [2,6]. For instance, adjuvant radiotherapy has demonstrated potential in reducing 5-year local recurrence rates in spermatic cord sarcomas with close margins [6]. Similar findings have been pinpointed in other rare para-testicular sarcomas, including leiomyosarcoma, where aggressive local control with adjuvant therapy has been associated with reduced recurrence [1,4]. Despite this, the selection of appropriate patients for adjuvant therapy and the optimal regimens require further investigation [3].

Gaps in Current Management and Reporting

Several inherent challenges and gaps exist in the current understanding and management of DDLPS of the spermatic cord:

- **Diagnostic Ambiguity**: The non-specific clinical presentation of a painless inguinal or scrotal mass often leads to misdiagnosis, as it can mimic benign conditions like hernias or epididymal cysts [6]. While imaging provides valuable information on mass characteristics, it frequently lacks the specificity required for definitive differentiation between benign and malignant lesions, necessitating histopathological confirmation [6]. Diagnostic challenges of similar nature have been highlighted in other liposarcoma and leiomyosarcoma case reports, where preoperative imaging often failed to distinguish malignancy [4,5]. Our case, where the tumour was identified incidentally during surgery for a suspected hernia, reflects this common diagnostic pitfall.
- **Lack of Standardized Guidelines**: The extreme rarity of this malignancy translates to a scarcity of level one evidence-based standardized treatment algorithms and official guidelines for diagnosis, treatment, and follow-up [2,6]. Current management strategies largely rely on data extrapolated from other soft tissue sarcomas, case reports, and single-institution experiences [2,3].
- **Importance of Multidisciplinary Approach**: The complexity of DDLPS necessitates a multidisciplinary approach involving surgeons, pathologists, oncologists, and radiologists to ensure accurate diagnosis and comprehensive treatment planning [3,6].
- **Follow-up Duration**: The present case report describes the patient's postoperative course as uneventful and likely includes a limited follow-up period, similar to many single case reports. However, due to the high risk of

recurrence and metastatic potential of DDLPS, prolonged surveillance with physical examination and cross-sectional imaging is critically warranted [2,6]. A one-year follow-up period, as often seen in case reports, is generally insufficient for a thorough assessment of long-term prognosis and recurrence rates in such aggressive malignancies [6]. Recent reports also emphasize the need for extended follow-up to capture late recurrences that may occur several years post-surgery [5].

CONCLUSION

To advance the understanding and improve outcomes for DDLPS of the spermatic cord, future research should focus on several key areas:

- **Multi-institutional Collaboration**: Given the rarity of DDLPS, collaborative international efforts are essential to aggregate clinico-pathological data. This would allow for larger datasets, enabling more robust analyses of prognostic factors, treatment efficacy, and long-term outcomes, thereby overcoming the limitations of single-case reports and small institutional studies [3].
- **Standardization of Diagnostic and Treatment Protocols**: Research is needed to develop evidence-based standardized guidelines for diagnosis, pathological assessment (including routine MDM2 and CDK4 testing), and treatment of DDLPS of the spermatic cord. This includes defining the utility of preoperative biopsy, which currently lacks sufficient sensitivity and specificity data [2].
- **Optimization of Adjuvant Therapies**: Further studies are required to elucidate the optimal role of adjuvant radiotherapy and chemotherapy, including appropriate patient selection criteria and effective regimens for high-risk DDLPS.
- **Long-term Follow-up Studies**: Prospective studies with extended follow-up periods are crucial to better understand recurrence patterns, metastatic potential, and long-term survival rates, ultimately leading to more refined surveillance strategies.
- **Molecular Characterization**: Deeper molecular characterization of DDLPS of the spermatic cord could identify novel therapeutic targets and contribute to the development of individualized treatment approaches [6].

This case report contributes valuable information to the sparse literature on DDLPS of the spermatic cord, emphasizing the need for a high index of suspicion, meticulous diagnosis, and aggressive surgical management. It also highlights the critical need for further collaborative research to establish definitive guidelines and improve patient outcomes for this challenging disease.

Authors' Contributions

We certify that we have participated sufficiently in the intellectual content, conception and design of this work or the analysis and interpretation of the data [when applicable], as well as the writing of the manuscript, to take public responsibility for it and have agreed to have our name lister as a contributor.

Declaration by Authors

Ethical Approval: The authors certify that all appropriate patient consent forms in accordance with the Declaration of Helsinki were duly obtained and is archived. The patient has given consent to publish the images and other clinical information in the journal. Care was taken to maintain the anonymity throughout the report.

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Conflict of Interest: Authors declare no conflict of interest.

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