CASE REPORT OPEN ACCESS

Surgical Management of Testicular Sarcoma: A Case Report

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ABSTRACT

The aim was to present a case report depicting the gradual onset of symptomatic testicular sarcoma. It was histopathologically proven as sarcoma and a small component of conventional seminoma. The patient presented to us with complaints of left testicular swelling, abdominal pain, and weight loss. The patient was treated with a left radical orchidectomy and left double J stenting. Sarcomas encompass a diverse array of malignant tumors arising from mesenchymal tissues, characterized by their rarity. They constitute less than 1% of all malignancies in adults and approximately 12% in pediatric populations. Most primary testicular tumors originate from germ cells. Testicular sarcoma has a good prognosis after surgical resection of localized tumors, while patients with distant metastasis have a poorer prognosis. Treatment option includes radical orchidectomy and post-operative chemotherapy.

Keywords: Testicular, Sarcoma, Orchidectomy, Stenting, Seminoma, Germ Cell.

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INTRODUCTION

Testicular cancer represents 1% of adult male neoplasms and 12% of pediatric malignancies [1] and is a relatively rare disease[2]. Sarcomas have a mesenchymal origin and are categorized into heterogeneous malignant tumors. Their distribution is at various sites in the body, extremities being the major component. A study that included 4550 adults with soft tissue sarcoma had different sites: 46% of these tumors occurred in the thigh, buttock, and groin area, with 13% appearing in the upper extremities. Torso and retroperitoneum combined accounted for 18%, while head and neck locations constituted only 9%. Treatment for sarcomas varies depending on the site and histology[1].

Intra-scrotal sarcomas are divided into para-testicular and intratesticular tumors.

Para-testicular tumors mostly include rhabdomyosarcomas in children and occasionally occur in older men[3]. Due to the potential for lymph node dissemination and early hematogenous spread, rhabdomyosarcomas have a typically aggressive course. The treatment protocol typically involves adjuvant chemotherapy and retroperitoneal lymph node dissection. Additionally, retroperitoneal radiation may be administered in cases where lymph node spread has occurred [3].

Para-testicular tumors, which occur in older men include leiomyosarcoma,fibrosarcoma, liposarcoma, and rhabdomyosarcoma, all of which comprise heterogeneous groups of tumors originating from the supportive and para-testicular structures [3]. Para-testicular tumors typically present without other components, unlike intratesticular tumors, which may be associated with germ cell tumors. Occasionally, both gonadal and extragonadal germ cell tumors undergo sarcomatous transformation. These sarcomas can arise spontaneously or in patients who have

undergone chemotherapy. The tumors encompass rhabdomyosarcoma, leiomyosarcoma, angiosarcoma, and undifferentiated types[3]. As reported by a study done in 1946 spermatolytic seminoma was first described as a separated histologic variant, we present a case with similar histopathological findings of a sarcoma and small component conventional seminoma [4].

CLINICAL PRESENTATION

A 50-year-old male, smoker, married, with no co-morbid presented to us with complains of: left testicular swelling for 5 months (gradually increasing with time) On and off abdominal pain, decreased appetite and weight loss for 3 months. On inspection, large swelling noted in left hemi scrotum with normal over lying skin. Palpation revealed swelling, which was size of a tennis ball, hard, non-tender, and smooth in consistency. The cord was palpable above the swelling, transillumination and cough impulse were negative. Inguinal lymph nodes were not palpable.

Ultrasound testis showed enlarged left testis with evidence of a large heterogeneous mass measuring 6.0x4.3 cm in the left testicular parenchyma. The mass was almost replacing normal testicular parenchyma.

Ultrasound abdomen showed a heterogenous mass measuring 8.0x6.2 cm seen at left abdomen probably nodal mass and causing a pressure effect over left ureter, and causing mild hydronephrosis and hydroureter. CT chest and abdomen revealed multiple heterogeneously enhancing hypodense nodal deposits in para-aortic region, the largest measuring approximately 62 x 63 mm in AP and TS diameter, it was identified at the level of the left renal pelvis, completely encasing the left aorta, resulting in its mild compression and displacement. No definite invasion seen. It was causing a compressive effect over the left renal pelvis, resulting in mild left-sided hydronephrosis as seen in **Figure 1**.

Posteriorly, it is closely abutting the left psoas muscle. These were extending along left common iliac vessel up to its mid part. The largest left common iliac lymph node measured 30 x 22 mm. Few well-defined nodular densities were seen scattered in bilateral lung fields, few of them were in subpleural location. The largest nodular density in left lung measured 6 x 5 mm was identified in superior basal segment of left lower lobe, the largest nodular density measured 7 x 6 mm was identified along the lateral segment of right middle lobe in subpleural location. Possibility of pulmonary metastasis. The tumor markers were in normal range. Radical orchidectomy was planned followed by left sided DJ stenting.

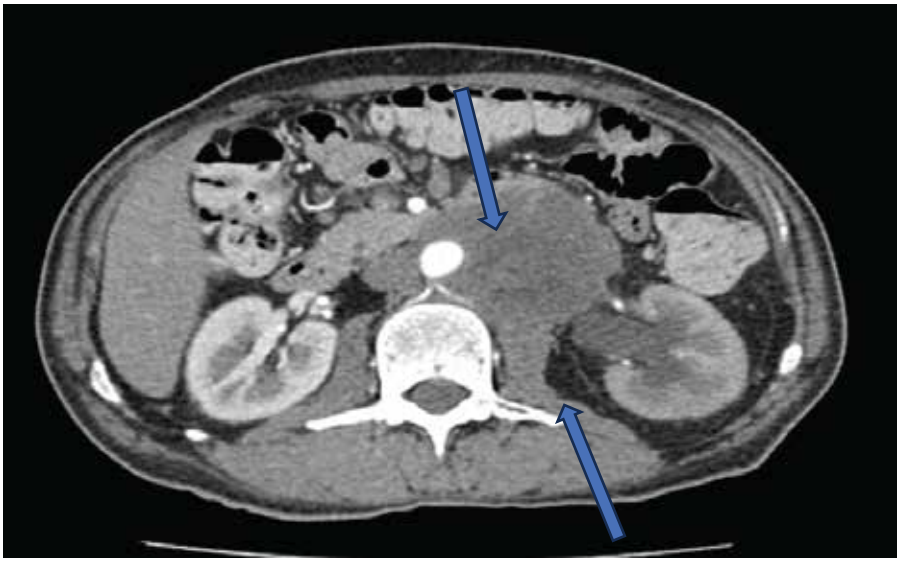

Figure 1: **Heterogeneously Enhancing Hypodense Nodal Deposits in The Para-Aortic Region, The Largest Measuring Approximately 62 X 63 Mm in AP and TS Diameter, It Was Identified at The Level of The Left Renal Pelvis, Completely Encasing the Left Aorta Resulting in Its Mild Compression and Displacement Causing a Compressive Effect Over the Left Renal Pelvis Resulting in Mild Left-Sided Hydronephrosis.**

Histopathology revealed testicular sarcoma, consisting of poorly differentiated sarcoma and a small component of germ cell tumour. There was extensive necrosis in tumour compromising morphologic evaluation. Focal areas showed lobules of necrotic cells with rounded nuclei shown in **Figure 2.**

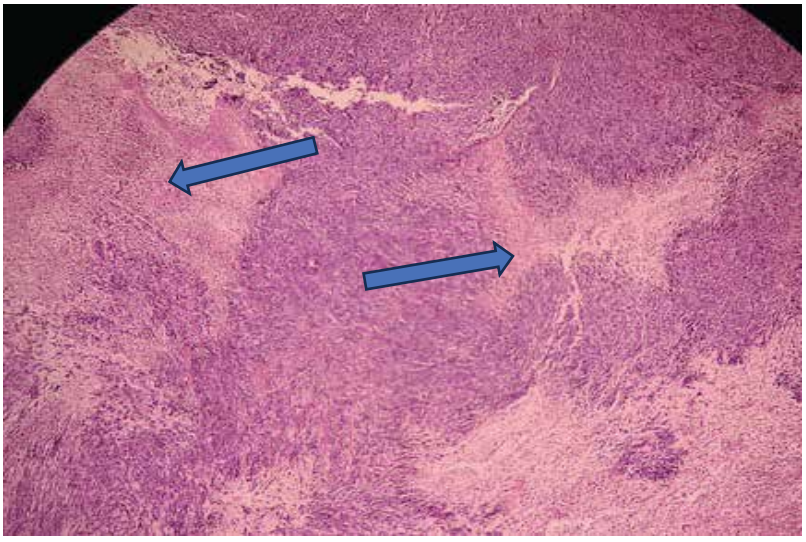

Figure 2: **Spindle Cells with Large Areas of Necrosis. Magnification Power 4X.**

Filamentous chromatin raising a possibility of a component of spermatocytic tumour. The tumor cells exhibited large vesicular nuclei, prominent nucleoli, and moderate eosinophilic cytoplasm, accompanied by numerous mitotic figures, as illustrated in **Fig 3.** The tumor was labeled was T4N2M1A. International consult was taken from Dr Christopher D.M Fletcher, and it was concluded that there is undifferentiated malignant epithelioid and spindle cell neoplasm, representing sarcoma and small component conventional seminoma.

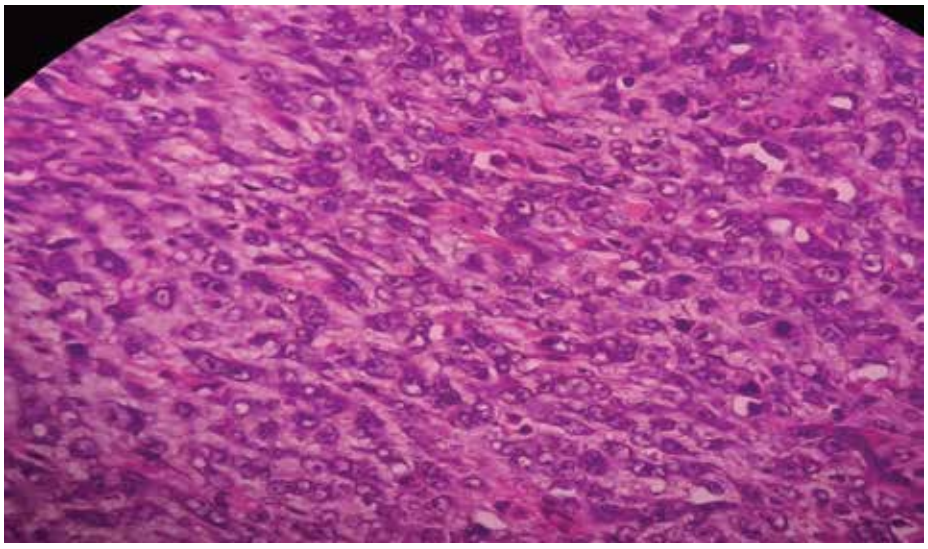

Figure 3: **The Tumor Cells Exhibited Large Vesicular Nuclei, Prominent Nucleoli, and Moderate Eosinophilic Cytoplasm. Numerous Mitoses Seen. Magnification Power 40X.**

DISCUSSION

Sarcoma is one of the significant types of malignant proliferation of cells. Sarcoma itself is a broad term for cancers that originate in connective tissue, such as bones and soft tissues of the body, which makes prevalence anatomically substantial. Additionally, soft tissue sarcoma targets structures that connect, support, and surround other viscera [5].

There are more than 70 types of sarcomas categorized by type, prevalence, and relative management. Adult testicular sarcoma arises from scrotal tissue and extends through epididymis, spermatic cord, and tunica vaginalis [5].

Testicular cancers can result from both modifiable and non-modifiable risk factors. Non-modifiable risk factors include age, which is around 20-34 years however can be seen at the age of 40 and above, cryptorchidism, genetic inheritance i.e Klinefelter

syndrome [6]and race; it is 4-5 times more prevalent in Caucasian men[7]. Modifiable risk factors include tobacco smoking, acquired immunodeficiency syndrome, in synchronization with other risk factors for example infrequent exposure to human papillo-ma virus [8], exposure to radiation and previous history of testicular sarcoma.

Often, all testicular cancers present as a painless mass, which can be painful and is associated with swelling and hardness in the scrotum. Patients with testicular lumps undergo testicular ultrasonography and MRI5. 5% of these patients may exhibit metastatic disease, presenting with symptoms such as a neck or abdominal mass, lumbar back pain, cough, hemoptysis, dyspnea, or gastrointestinal symptoms. Additionally, gynecomastia occurs in nearly 10% of men with testicular cancer due to tumors that secrete (βHCG)[9].

Evaluation of nodal and distant metastasis can be done with the use of PET /CT5. **Figure 1** shows pulmonary metastasis. **Figure 2** shows abdominal mass mostly lymph nodes encasing aorta and causing pressure effect. The mass is completely encasing left ureter causing left sided hydroureter.

A study conducted in 2021, examining the clinical characteristics and prognostic factors of testicular sarcoma. Their research, based on registry data from 158 testicular sarcoma patients, revealed that all individuals, with a median age of 17.00 years (ranging from 1.00 to 93.00 years), were pathologically diagnosed through orchiectomy or needle biopsy specimens.Among the patients studied, there were 42 (30.43%) cases classified as Tis, 53 (38.41%) as T1, 15 (10.87%) as T2, 20 (14.49%) as T3, 5 (3.62%) as T4, and 3 (2.17%) cases where the invasion degree exceeded the staging system of testicular cancer. Localized metastasis occurred in 31 (20.13%) patients, while distant metastasis was found in 28 (18.18%) cases. A majority, 61.69% (n = 95), showed no signs of metastasis. Regrettably, 32 (20.25%) patients succumbed to the disease. The study revealed that patients with distant metastasis were more likely to pass away within one year of diagnosis[1].

The TNMS staging system, which considers primary tumor extent, regional lymph node involvement, presence of distant metastasis, and serum tumor markers, was established by the American Joint Committee on Cancer. Staging determines the amount of The spread of the primary tumor within the tissue of the testicle, its extension to regional lymph nodes, metastasis to other organs, and the presence of serum tumor markers are key considerations in assessing the stage and prognosis of testicular cancer[9].

The primary treatment for testicular cancers is typically surgery, aiming to completely remove the tumor with negative margins. The prognosis of testicular sarcoma depends on several factors including stage, grade, and the extent of surgical resection. In cases of high-grade tumors with positive margins, there are additional considerations such as adjuvant chemotherapy, radiation therapy, and additional surgical interventions like retroperitoneal lymph node dissection to reduce the risk of recurrence[7].

In certain cases when wide resection of the tumor was not possible because of microscopic tumor seeding or comorbidities associated with the patient, adjuvant radiation therapy was considered to control local spread[5].

Primary testicular sarcoma being a rare tumor, it is difficult to establish categorical treatment recommendations. Inguinal orchiectomy is the first line surgery performed, followed with careful surveillance with BHCG and AFP. With the aid of these serological tumor markers, while not specific to sarcoma, we can rule out serologic relapse as a sign of undetected germ cell tumor metastases. It's worth noting that less than 5% of all histologic soft tissue sarcomas demonstrate lymphatic metastases, whereas hematogenous spread is observed in adjacent genital organs, spermatic cord sarcomas and scrotal wall sarcomas. On the contrary germ cell tumors have retroperitoneal nodal metastases [10].

Neoadjuvant chemotherapy offers several advantages, including tumor downsizing, facilitating less extensive surgical excisions, and assessing chemotherapy sensitivity. However, its primary significance lies in its ability to target micro metastases and potentially lower the risk of metastatic disease development. Although preoperative chemotherapy is increasingly employed in many centers for patients at high risk of metastatic disease, based on factors such as sarcoma histopathology, tumor size, and grade, the utilization of neoadjuvant chemotherapy still varies depending on the institution.

Extensive studies have been conducted to investigate the role of adjuvant chemotherapy in sarcomas. Another study conducted a meta-analysis which was published in 1997, it provided compelling evidence of a survival benefit, showing an 11% absolute risk reduction in mortality[5].

CONCLUSION

Testicular sarcoma is one of the rarest carcinomas in the world with less than 1% of total carcinomas. However, they have a good prognosis after surgical resection (radical orchidectomy). Patients with distant metastasis have a poor outcome, with a

survival of less than one year. Treatment options include radical orchidectomy and post-surgical chemotherapy.

CONFLICT OF INTEREST

None

AUTHORS' CONTRIBUTIONS

AHJ: Conceived the idea and identified the case and critically revised the manuscript, **MAA** collected clinical data and literature searches, **ZS** and KI wrote the initial draft and managed the literature searches, **YM:** reviewed the literature. All authors approved the final version for submission.

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