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Massive growth of paratesticular myxoid liposarcoma recurrence and inguinal metastases following scrotal violation: A case report

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ABSTRACT

Paratesticular myxoid liposarcomas are exceedingly uncommon soft tissue tumor, comprising merely 3.3% of all liposarcomas in paratesticle. The primary treatment involves total surgical removal with orchiectomy performed through an inguinal incision. Even though radical orchiectomy is still the best treatment to date, recurrence is still inevitable. There is possibility of tumor recurrence and metastasis due to scrotal violation. Trans-scrotal approach of orchiectomy increased the possibility of local recurrence and possible spread by allowing the cancerous cells to leak into the adjacent tissues. We discuss the clinical presentation of the case, the therapeutic management, and the possible complications after scrotal violation of prior removal surgery. We report a case of a 69-year-old man with painful recurrent scrotal tumor along with massive growth and spread to the spermatic cord and inguinal region after right orchiectomy with trans scrotal approach one year earlier. The tumors were 23 x 25 x 8 cm for the inguinal tumor, 13 x 9 x 7 cm with ulcerative surface for the scrotal tumor, and 12 x 6.5 x 5 cm for the spermatic cord tumor. Our patient then had wide excision tumor surgery with a radical lymphadenectomy. Similar cases of recurrent paratesticular myxoid liposarcoma have been reported; however, the recurrence occurred over a year and no metastases had been found. In our case, the recurrent tumor had significant development rapidly within a year after scrotal violation and was also found to have already metastasized to the inguinal region. Massive recurrence tumor growth and spread in paratesticular liposarcoma following orchiectomy is rarely heard of. Surgical treatment is still the preferred treatment to date, but local recurrence and metastasise somehow is inevitable. Tumor recurrence and spread hopefully can be prevented by proper diagnosis and surgical management of the tumors; high ligation of the cord, inguinal approach and ensure negative margin of tumors. This case report also highlights the significance of avoiding scrotal violations to prevent tumor seeding to adjacent sites, thereby reducing patient morbidity and mortality.

Keywords: *Myxoid liposarcoma, paratesticular tumor, scrotal violation, scrotal mass*

1. INTRODUCTION

Paratesticular neoplasms are extremely rare; they make up 7% of all intra-scrotal tumors, and among those tumors, 5% are liposarcomas, which are typically present in older adults.[1,2] Based on the WHO 2020 classification of soft tissue tumors, liposarcomas are categorized into: well- differentiated, dedifferentiated, yxoid, pleomorphic, and myxoid pleomorphic.[3] Paratesticular myxoid liposarcomas are uncommon, comprising merely 3.3% of all liposarcomas in this anatomical site. These tumors typically manifest between the fourth and fifth decade of life.[4,5] However, often, this entity misdiagnosed as cord lipoma, inguinal hernia, or hydrocele. The lack of a standardized treatment pathway for paratesticular liposarcoma also makes managing paratesticular myxoid liposarcoma challenging, even though most cases were treated with wide excision and possibly orchiectomy.[6]

Around 5--30% of liposarcoma manifest as myxoid liposarcoma with predominant localization to extremities.[7,8] This type of liposarcoma tends to metastasize irrespective of its grade, especially to soft tissue sites, even though a report shows a metastasis rate between 0,6—7%.[7] As stated by several case reports, a common problem faced by patients with para testicular myxoid liposarcoma is the diagnosis delay; clinicians are unaware of this entity, and sometimes, suspicion of another etiology was raised if there were symptom recurrence.[6,9] Its aggressive nature makes its high recurrence or metastasis rate; a report showed a 22% metastasis rate with a median follow-up time of 7,6 years.[10] Several risks of developing metastasis and recurrence have been reported, including surgical procedures. The risk of seeding a metastasis after a biopsy procedure is very small.[11] Patients with marginal resection of myxoid liposarcoma showed a higher recurrence than wide surgical resection.[12] Another surgical issue is a scrotal violation, especially in testicular cancer, a

nonstandard surgical approach that includes scrotal orchiectomy, open testicular biopsy, and fine needle aspiration.[13] It was associated with a high recurrence rate but no metastasis or survival change in the context of testicular masses suspicious of malignancy.[14] However, the role of scrotal violation in the risk of recurrence and metastasis of paratesticular myxoid liposarcoma is still unknown.

We describe a rare case of paratesticular myxoid liposarcoma recurrence with massive growth and spread within a year after the initial scrotal tumor was completely removed. This condition is rarely heard of, as reports of a recurring tumor on an extensive level following total removal are still rare. Here we adhere to CARE guideline to present our case report.[15]

2. CASE REPORT

A 69-year-old man was admitted to our emergency department with painful massive tumor from the scrotum up to the right inguinal region, evolving in the past month before being admitted. He had history of painless right scrotal tumor and had been surgically removed through an incision made on the scrotum one year ago. The post operative histopathological result was suggestive of myxoid. Unfortunately, the tumor recurred along with significant growth and spread within a year after tumor removal.

Figure 2. Ulcerative surface of tumor recurrence following right orchiectomy

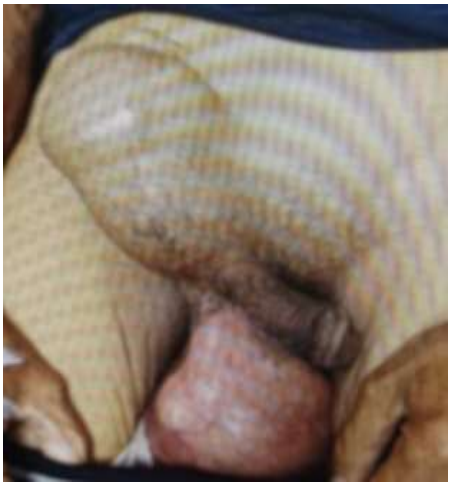
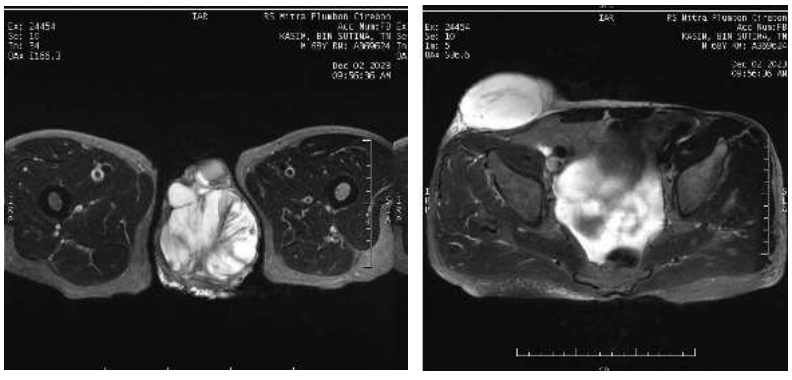


Figure 2. Massive tumor growth and metastasis from right paratesticular up to right inguinal region following right orchiectomy

The right inguinal lump's initial diameter was one centimeter, but it grew extensively without noticeable stimulating or relieving factors. The tumor surface area was also ulcerative (Figure 1). All laboratory tests were normal. A tumor marker examination was not available in our center. There are no other signs or symptoms.

From the clinical examination of the tumors, we found an apparent tender mass in the scrotum area up to the inguinal region and ulcers on the inferior part of the testicle measuring 10 x 8 x 6 cm (Figure 2). Based on the MRI result, malignant masses in the right intrascrotal with infiltration of the scrotum skin layer that forms ulcers and spread to the inguinal canal, malignant characteristic of lymphadenopathy at the right inguinal (non-regional), and ascites in perivesical (Figure 3). \



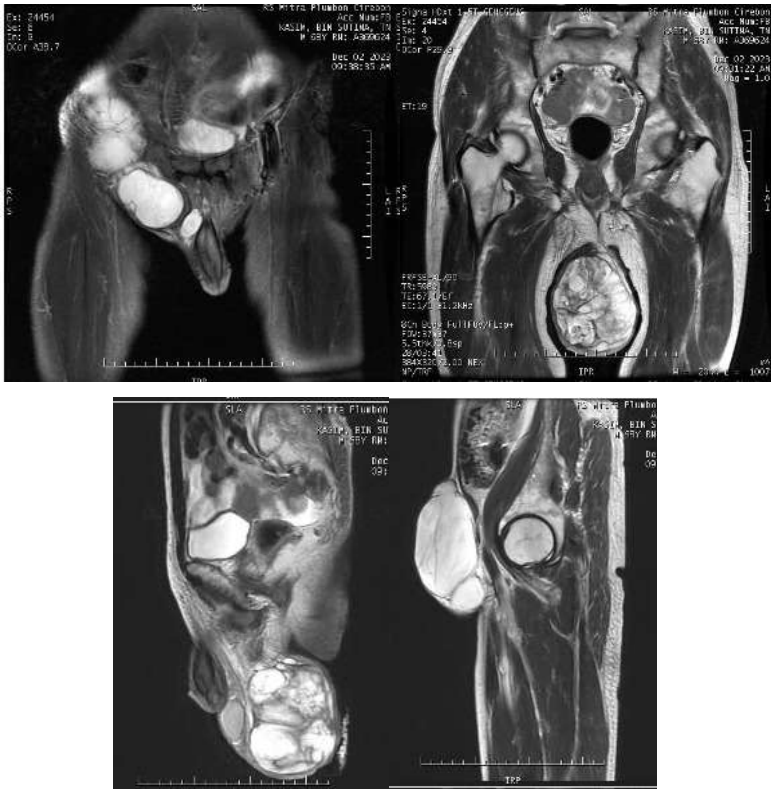


Figure 3. Abdominal pelvis MRI shows malignant masses in the right intrascrotal with infiltration of the scrotum skin layer and malignant lymphadenopathy at the right inguinal

This patient underwent tumor-wide excision with radical lymphadenectomy of the adjacent lymph nodes. The size of the inguinal tumor was 23 x 25 x 8 cm, the scrotal tumor was 13 x 9 x 7 cm, and the spermatic cord tumor was 12 x 6.5 x 5 cm. The post-operative histopathological examination revealed that the tumor's cell growth was diffuse, pleomorphic, hyperchromatic, and mitotic; no metastasis to the adjacent lymph node had been found. The tumor was diagnosed with high-grade myxoid liposarcoma. The recovery after the surgery was uneventful.

The patient came to our Urology clinic without complaint one month after surgery, and the wound recovered well. There was no evidence of tumor recurrence, but the patient will have regular follow-up every 6 months to have a thorough examination for the possibility of tumor spread or recurrence.

3. DISCUSSION

Genitourinary sarcoma is a rare kind of malignancy, accounting for around 1%–2% of all malignant tumors within this region.[16] Liposarcoma is the second most prevalent soft tissue tumor in adults, originating from primitive mesenchymal cells in adipose tissue.[1,17] Paratesticular liposarcoma represents fewer than 12% of all liposarcoma cases. Around 90% of paratesticular liposarcomas originate from the spermatic cord, with the remaining cases arising from the testis tunica vaginalis and epididymis.[18–20] Paratesticular myxoid liposarcomas comprise merely 3.3% of all liposarcomas in this anatomical site.[4,5]

There is a similar case of recurrent paratesticular liposarcoma after tumor resection that was also reported by Hua R et al.; the initial size was 22 cm × 14 cm × 8 cm and recurred later to the size of 26 cm × 17 cm × 13 cm 2 years after the resection.[18] Meanwhile, in our case, one year after the resection, the tumor already had massive growth and spread; the inguinal tumor was 23 x 25 x 8 cm, the scrotal tumor was 13 x 9 x 7 cm, and the spermatic cord tumor was 12 x 6.5 x 5 cm. The size was bigger than the case reported by Hua R et al., which had a shorter duration for tumor growth.

Paratesticular liposarcoma typically manifests as a gradual lump in the groin or scrotum, which tends to grow continuously without causing pain or discomfort.[18] Our patient comes with a painless lump that is initially 1 cm in size but gradually grows. Before the first surgery, the scrotal tumor was 21 x 17 x 4 cm in size. Because it is painless and slowly growing at first, the symptoms were overlooked and not considered, leading to a delay in treatment initiation until the tumor size got bigger.

Myxoid liposarcoma typically manifests as a painless soft tissue tumor, based on cases reported by Hua et al. and Hemalatha

et al.[5,16,18,19] This report was different from our case in that after recurrence, our patient's chief complaint was a painful tumor, which can be attributed to two main factors: the ulcerative surface due to infection and the substantial size of the tumors that strained the surrounding structure. Myxoid liposarcoma is also aggressive; a review by Qu et al. showed that 78% of metastatic myxoid liposarcoma is categorized as high-grade. The approximate five-year local recurrence and metastasis for myxoid liposarcoma are 6—13% and 13—25%, respectively.[7]

The simple diagnostic tool for paratesticular liposarcoma is ultrasonography, and it often reveals solid, hyperechoic, and diverse lesions. If the tumor was small, the differential diagnosis was probably hernia, testicular or epididymal tumor, hydrocele, and hematoma. However, determining the tumor's origin and boundaries is challenging for big tumors and requires an advanced examination. Laboratory biochemical assays are not specific enough for diagnosing paratesticular liposarcoma.[16,18]

CT scan and MRI can help identify the tumor's precise location, histology, and morphological characteristics and evaluate whether it has spread to adjacent tissue. The diagnostic examination of our patient is from the abdominal pelvic MRI and post-operative histopathological examination that is suggestive of myxoid liposarcoma. It is following the Hua et al case report stating that the diagnosis of paratesticular liposarcoma mostly relies on postoperative histopathology, immunohistochemical, and molecular testing. However, immunohistochemical and molecular testing are unavailable at our center, Gunung Jati Regional Hospital Cirebon, Indonesia. The decision for surgery was made based on the clinical and radiological exams that were feasible at that time.

The primary treatment for paratesticular liposarcoma involves total surgical removal with an orchiectomy performed through an inguinal incision.[19–22] This involves surgically removing the mass by excising it widely, which includes orchiectomy and high ligation of the spermatic cord to ensure a negative margin. According to Rodriguez et al., Ballo et al., Blitzer et al., and Sogani et al., the major primary surgical treatment involves complete removal of the tumor with radical inguinal orchiectomy and high ligation of the cord. [23–26] Chalouhy et al. also highlight the importance of aggressive surgical intervention, including broad resection of tumor recurrence, to reduce local recurrence and improve disease-free survival. [27] The most suggested treatment strategy by most authors is radical orchiectomy with high ligation of the cord and broad tumor excision, ensuring no microscopic positive margins.[28]

In this case, the initial scrotal tumor was taken without ensuring the negative tumor margin area. An incision was made through the scrotum. After the complete removal of the scrotal tumor, there was a recurrence with very rapid and massive growth and spread within a year. Even though radical orchiectomy is still the best treatment to date, recurrence is still inevitable.

Rei Kamitani et al. found that among 265 paratesticular liposarcoma patients analyzed, those who underwent high inguinal resection had a significantly greater recurrence-free survival rate than those with tumor resection. [18,29] Factors known to be independently associated with disease-specific survival in patients with decreased local recurrence rate after adjuvant therapies were tumor histology, negative margins after surgery, metastases, and tumor size.[30] A high local recurrence rate and a low distant recurrence rate characterize myxoid liposarcoma. It tends to be locally aggressive and tends to reappear after surgical removal.[19]

Tuzzato et al reported that approximately 12–25% of individuals with high-grade myxoid liposarcoma experience local relapses, whereas 30–60% develop pulmonary or extrapulmonary metastases within a 10-year follow-up period.[31] A local spread refers to the presence of a disease in the scrotal or inguinal regions and the superficial lymph nodes. The distant spread encompasses all additional locations, including biochemical recurrence indicated by increased serum tumor markers.[32,33]

In our case, the first surgery was not a radical orchiectomy; the spermatic cord and adjacent lymph glands remain intact, and the tumor margin was not confirmed negative in the histopathological exam. Treatment choices in this case were only surgical-wide excision; there was no adjuvant radiation or chemotherapy given to this patient. Dürr et al. found from 43 patients with myxoid liposarcoma, radiation therapy significantly reduced the size of tumors; however, it did not affect local recurrence.[34] The effects of adjuvant postoperative radiotherapy remain unclear. [2]

The highlight of this case was also suspicion of scrotal violation that was done in the first surgery, which could be one of the factors leading to the spread and rapid growth of the tumor. Scrotal violation refers to any surgery that penetrates the scrotum and can lead to the spread of tumors. The approach of the initial right orchiectomy was trans scrotal. Trans-scrotal orchiectomy was not a preferred method as it increased the possibility of local recurrence and possible lymphatic metastases by allowing the cancerous cells to leak into the scrotal skin. [21,22]

The preferred approach for the orchiectomy is the inguinal method instead of the scrotal one; the scientific rationale is to minimize disruption to the lymphatic drainage system, reducing the risk of disease dissemination.[32,33] In their meta-analysis of 1182 cases, Capelouto et al. found a significant difference in the local recurrence rates of testicular cancer between patients who had scrotal violation (2.9%) and those who had inguinal orchiectomies (0.4%).[33] It is acknowledged that liposarcoma can spread through the infiltration of the spermatic cord and soft tissues around the inguinal canal or blood metastasis. However, lymph node metastasis with scrotal violation is rare.[18,34,35] Individuals with myxoid liposarcomas

typically have a favorable prognosis. The 10-year overall survival rate was 72%.[34]

The limitation of our case report is the lack of immunohistochemical and molecular analysis examination because of the limited resources; these results provide a substantial finding and may influence the chosen therapy for this patient and inform the patient of the recurrence risk. We also do not conduct whole-body MRI as recommended in patients with a new diagnosis of myxoid liposarcoma. It may be incorporated into the patient's management to provide holistic care for the patient.

4. CONCLUSION

Massive recurrence of tumor growth and spread in paratesticular liposarcoma following orchiectomy is rarely heard of. Surgical treatment is still the preferred treatment to date, but local recurrence and metastasis are somehow inevitable. Tumor recurrence and spread hopefully can be prevented by proper diagnosis and surgical management of the tumors, high ligation of the cord, and ensuring a negative margin of tumors. This case report also highlights the significance of avoiding scrotal violations to prevent tumor seeding to adjacent sites, thereby reducing patient morbidity and mortality.

Ethics approval

Ethical approval was not required for this study.

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Competing interests

All the authors declare that there are no conflicts of interest.

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Underlying data

Derived data supporting the findings of this study are available from the corresponding author on request.

Declaration of artificial intelligence use

We hereby confirm that no artificial intelligence (AI) tools or methodologies were utilized at any stage of this study, including during data collection, analysis, visualization, or manuscript preparation. All work presented in this study was conducted manually by the authors without the assistance of AI-based tools or systems.

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