Case Report

# Management and Prevention of Recurrent Paratesticular Liposarcoma

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## Abstract

**Paratesticular liposarcoma is a rare entity. Compared to other genitourinary sarcoma, it is usually detected earlier and is easily resectable, resulting in an excellent prognosis. The recurrence of well-differentiated paratesticular liposarcoma after complete resection is extremely rare. Optimal management of the tumour recurrence includes complete re-excision and radiotherapy to the area of recurrence. Here, we describe a 48-year-old man with a recurrent left paratesticular well-differentiated liposarcoma, six years after its complete excision. Our discussion focused on the therapeutic strategy to prevent tumour recurrence. It is hoped that this case discussion can increase the awareness of this condition and assist in its management.**

***Keywords:*** *adult, genitourinary, liposarcoma, sarcoma*

## Introduction

Paratesticular soft tissue sarcomas (PTSTS) are rare. This type of tumours represents approximately 0.01–0.02% of all malignant tumours (1). Liposarcoma represents only a small subset of the family of PTSTS, and its incidence and recurrence rate is unknown. Due to the paucity of reported cases, there is no standardised guideline for the management of these types of tumours. Consequently, treatment recommendations are based on case reports, small series and literature reviews, resulting in a number of unsolved issues. Surgical excision is the mainstay of treatment, but the use of adjuvant therapy to prevent recurrence remains debatable. In this article, we present a case of recurrent well-differentiated paratesticular liposarcoma after its complete excision. Our discussion focused on its management and the therapeutic measures that can be taken to help reduce tumour recurrence.

## Case Report

A 48-year-old man presented with a painless left inguinoscrotal swelling, which was slowly increasing in size over a period of five months. He had undergone left orchidectomy and a complete excision of the left paratesticular liposarcoma via inguinal approach six years before and the histopathology was well-differentiated liposarcoma with a clear surgical margin. On follow-up, his health remained well and computed tomography (CT) of the thorax, abdomen and pelvis did not show any distant metastases.

The current physical examination showed a firm, painless mass in the left inguinoscrotal region that was 12 cm at its widest diameter. The transillumination test was negative. The right testis and spermatic cord were normal. No inguinal lymphadenopathy was detected. Ultrasonography showed an 8.8 × 4.9 cm subcutaneous tumour with fat density. He was diagnosed with left inguinoscrotal recurrent liposarcoma.

Later, through an inguinoscrotal incision, the tumour was excised completely and, after the resection, the surgical field was washed with distilled water. Intra-operatively, there was a multilobulated mass extending from the inguinoscrotal region to the thigh involving the adductor longus muscle, medially.

Histopathological examination of the tumour showed encapsulated lobules of fibrofatty tissue with varying sizes of adipocytes and occasional lipoblasts. There were a few scattered large, pleomorphic nuclei cells with coarse chromatin,

  

and occasional multinucleation. Occasional mitotic activity was seen. Immunohistochemically, the tumour stained positive for S100 but negative for desmin and actin. Based on the identical histopathology and immunohistochemistry of the current and previous tumour, we diagnosed the patient with recurrent well-differentiated paratesticular liposarcoma.

## Discussion

Paratesticular liposarcoma is a rare neoplasm. Most of the available information on these tumours is derived from small series or case reports. The largest series, with 131 cases of adult genitourinary sarcomas, was collected at the Memorial Sloan-Kettering Cancer Center between July 1977 and July 2003. Only 25 cases of paratesticular liposarcoma were reported; at the time of diagnosis, the median age of the patients was 55 years old (2). According to the 2002 World Health Organization's (WHO) histological classification of tumours, liposarcoma can be divided into the following five categories: atypical lipomatous tumour/well-differentiated; dedifferentiated; myxoid/ round cell; pleomorphic; and mixed-type liposarcoma. The well-differentiated variant is typically indolent.

The mainstay of treatment for paratesticular liposarcoma is complete surgical excision with orchidectomy through inguinal incision (1,3). Trans-scrotal orchidectomy is not encouraged because it risks spillage of the neoplastic cells in the scrotal skin, thus increasing the risk of local recurrence and potential lymphatic metastases. A less than 10% risk of recurrence was found when a wide local excision with a margin of 10 mm or greater was used (3). Liposarcoma tends to spread primarily through local extension. Lymphatic spread is uncommon and usually presents late in the course of disease. According to Fong et al., in a prospective study of 1772 sarcoma patients, the overall prevalence of lymph node metastasis was 2.6%. They concluded that elective lymphadenectomy is not indicated and it does not have any therapeutic advantage in the treatment of primary disease (4).

Liposarcoma is the most radiosensitive soft tissue sarcoma and a well-differentiated tumour responds better than other tumour subtypes (5). Surgical resection with a margin less than 10 mm, or cases involving residual tumour cells, will benefit from radiotherapy and help in the prevention of local recurrence. Radiation therapy is recommended as the sole therapy if the tumour is not resectable secondary to organ or blood vessel dissemination (6). Doxorubicin-based adjuvant chemotherapy has been shown to significantly improve the time to local and distant recurrence and overall recurrence-free survival in a meta-analysis of 14 randomised clinical trials of various localised resectable soft tissue sarcoma. However, for liposarcoma the value of systemic therapy remains controversial (7).

The risk factors for developing local recurrence include the presence of a high grade tumour, large tumour size (> 5 cm), and inadvertent intralesional surgery. Repeat wide local excision of the tumour in local recurrence remains the cornerstone of treatment. To control regional recurrence, wide en bloc excision of all potentially contaminated tissue is important (8). Distilled water lavage after tumour resection minimises contamination and reduces tumour cell 'spillage'. Due to its hypotonicity, distilled water lyses free tumour cells and has a superior tumouricidal effect when compared to normal saline (9). Adjuvant radiation therapy post-surgery is recommended to reduce the locoregional failure. Fagundes et al., reported that patients who underwent surgical resection and adjuvant radiotherapy may effectively control the locoregional microscopic disease (10).

In summary, recurrent well-differentiated paratesticular liposarcoma is best managed by using a wide local excision via inguinal approach. Lymphadenectomy is not recommended. Distilled water should be used to lavage the surgical field. It was found that adjuvant radiotherapy reduced local tumour recurrence while adjuvant chemotherapy is controversial.

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