

Case Report

# Dedifferentiated Liposarcoma in the Spermatic Cord Finally Diagnosed at 7th Resection of Recurrence: A Case Report and Bibliographic Consideration

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## Keywords

Recurrence · Sarcoma

## Abstract

Liposarcoma in the spermatic cord is infrequent, and accurate diagnosis of histopathological subtype is often difficult in spite of the importance of differential diagnosis for adequate treatment. A 54-year-old man underwent left-sided high orchiectomy with inguinal lymphadenectomy for a spermatic cord tumor in July 2006. The initial histopathological report diagnosed leiomyosarcoma in the spermatic cord. He then underwent surgeries for repeated recurrences a further 6 times between July 2008 and May 2014. Pathological finding at the 7th resection of the recurrent tumor was osteosarcoma, which was uncommon in the sper-

matic cord. With a thorough overview of all specimens, the histopathological diagnosis was finally confirmed as dedifferentiated liposarcoma because of a biphasic pattern in the specimen of high orchiectomy at the first resection. A biphasic pattern represents high-grade sarcoma like osteosarcoma and well-differentiated liposarcoma, and is characteristic of dedifferentiated liposarcoma. Although the dedifferentiated type is one of poor prognosis, the diagnosing of liposarcoma histopathologically was found to be difficult throughout this case. In this report we discuss the accurate histopathological diagnosis of liposarcoma in the spermatic cord in order to prevent repeated recurrences based on a review of the literature, as well as the difficulty in recognizing dedifferentiated liposarcoma macroscopically and morphologically. Our experience suggests that, after much difficulty, accurate histopathological diagnosis of liposarcoma in the spermatic cord is still clinically challenging.

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Published by S. Karger AG, Basel

## Introduction

Liposarcoma in the spermatic cord is comparatively infrequent [1]. Dedifferentiated liposarcoma is one of the subtypes with poor prognosis and the pathological finding of a biphasic pattern is characteristic [2, 3]. However, accurate diagnosis of histopathological subtype is often found to be difficult in spite of the importance of differential diagnosis throughout our experience in this case. This is because a tiny component of high-grade sarcoma, which is one of the pathological features of a biphasic pattern, is likely to be missed. Accurate diagnosis of histopathological subtype is needed for liposarcoma in the spermatic cord to prevent repeat recurrence.

## Case Report

A 54-year-old man noticed a painless mass in the left scrotum and visited our hospital in July 2006. Magnetic resonance imaging (T1- and T2-weighted) of the pelvis demonstrated a 30-mm hypointense mass in the left spermatic cord and swelling of a left inguinal lymph node (Fig. 1a) with no evidence of local invasion. Left high orchiectomy with inguinal lymphadenectomy was performed in July 2006. According to immunostaining examination, pathological diagnosis was leiomyosarcoma with actin-positive cells, and all margins were negative. Thus, no further treatment was performed.

During the 8-year follow-up, he underwent surgery 3 times for local recurrences (Fig. 1b–e) and 3 times for retroperitoneal metastases (Fig. 1f–h) (Table 1). The first local recurrence was identified as leiomyosarcoma in July 2008 with similar histopathological findings to those of the primary tumor. However, at the 2nd to the 5th recurrence surgically resected specimens were diagnosed as rhabdomyosarcoma (Fig. 2d), because a rhabdomyoblastoma with myoglobin-positive immunostaining cells was detected (Fig. 2e). At the 6th recurrence in January 2014 the surgically resected specimen had pathological findings of osteosarcoma (Fig. 2f). Reexamination of the specimen for the primary tumor revealed osteosarcoma without osteoid formation (Fig. 2a) and lipoblasts with spindle-shaped tumor cells (Fig. 2c).

These findings represented a biphasic pattern, characteristic of dedifferentiated liposarcoma (Fig. 2b). The final diagnosis was confirmed as dedifferentiated liposarcoma in the spermatic cord. After the final diagnosis, the patient underwent further surgery 3 times for local recurrences. Chemotherapy with Adriamycin following eribulin was performed in the hope of suppressing further progression. At 8 months after the final chemotherapy, the most recent CT scan revealed stable disease with local recurrences.

## Discussion

We encountered a case of liposarcoma of the spermatic cord that was finally diagnosed as dedifferentiated type after several recurrences, although the initial diagnosis had been leiomyosarcoma. Liposarcoma is generally classified into 5 subtypes according to the World Health Organization (WHO) classification: well-differentiated, dedifferentiated, myxoid, pleomorphic, and mixed. Since the probability of recurrence differs depending on subtype, precise diagnosis is important for dedifferentiated liposarcoma characterized by a biphasic pattern of high-grade sarcoma (including osteosarcoma without osteoid formation) and well-differentiated liposarcoma (including lipoblasts with spindle-shaped tumor cells).

In this case of leiomyosarcoma of the spermatic cord at first surgery, we initially missed a tiny component of high-grade sarcoma, due to our predisposition to depend too much on immunostaining examination in confirming the findings of hematoxylin and eosin staining. At the 7th surgery, it was strange to be informed that the pathological finding was osteosarcoma, which is not supposed to derive from the spermatic cord. To reach accurate diagnosis, we needed a thorough overview of every histopathological finding of the entire series of specimens.

While reviewing the primary tumor specimen from 2006, lipoblasts and spindle-shaped tumor cells characteristic of well-differentiated liposarcoma were observed, along with osteosarcoma without osteoid formation (Fig. 2a). These findings represent a biphasic pattern of osteosarcoma and well-differentiated liposarcoma (Fig. 2b, c), a finding characteristic of dedifferentiated liposarcoma. We eventually realized that a crucial part of the lipoblastoma in the specimen was initially missed from the high orchiectomy in 2006, because it was a tiny proportion of the overall mass. We may have misevaluated the specimen as leiomyosarcoma due to the prominent actin-positive appearance on immunostaining examination.

To our knowledge, a total of 326 cases, including ours, with spermatic cord liposarcomas have been reported and approximately a quarter of them were from Japan (online suppl. table; for all online suppl. material, see [www.karger.com/doi/10.1159/000479364](http://www.karger.com/doi/10.1159/000479364)). We reviewed all the cases to evaluate treatment strategies for spermatic cord liposarcoma. When divided into subtypes of liposarcomas according to the WHO classification, 146 cases were classified as well-differentiated (45.3%), 50 as dedifferentiated (15.2%), 34 as myxoid (10.6%), 5 as pleomorphic (1.6%), 14 as mixed (4.3%), and 77 as unknown (online suppl. table). Overall, 32 cases (9.9%) had recurrence, described in detail as 14/146 (9.6%) well-differentiated, 10/50 (20.0%) dedifferentiated, 5/34 (14.7%) myxoid, 1/5 (20%) pleomorphic, and 1/14 (7.1%) mixed, respectively. The patients with dedifferentiated or pleomorphic type showed a relatively high recurrence rate compared to those with other types,

suggesting that the first treatment is very important in order to prevent local recurrences and metastases.

It is reported that the recurrence rate of spermatic cord liposarcoma is approximately 10%. According to the previous 235 reports whose operations were described, high orchiectomies were performed in 214 cases and simple mastectomies in 20 cases. The recurrence rate was 9.8% (21/214) for high orchiectomy patients and 55% (11/20) in simple mastectomy patients, indicating that the recurrent rate was significantly higher in simple mastectomy patients. It has been shown in some reports that extended excision has a better prognosis, but it is still controversial because excessive extended excision requires resection of major blood vessels and organs around the spermatic cord, leading to a deterioration of quality of life of patients.

The Federation Nationale des Centres de Lutte contre le Cancer (FNCLCC) grading system has been used to evaluate the malignancy of soft-tissue sarcomas. This grading scores sarcomas according to tumor differentiation, mitotic count, and necrosis, and provides a final evaluation score between grade 1 and grade 3. Accurate histopathological diagnosis is important, because significant differences in the overall survival rate are seen among grades 1, 2, and 3. The dedifferentiated type classified as grade 2 or 3 is one of the types with poor prognosis.

A wide and complete resection with negative microscopic margin is crucial to surgery of liposarcoma in the spermatic cord. There is no standard management except for surgery [4]. With regard to the effectiveness of adjuvant therapy [5, 6], the addition of radiotherapy (50–60 Gy) is efficacious in the case of resected margins within 1 cm [7]. Moreover, pre- and postoperative chemotherapy with Adriamycin, ifosfamide, and eliburin reportedly improves prognosis for grade 2 or 3 sarcomas of more than 5 cm [8]. Hence, adjuvant chemotherapy and/or radiotherapy should be considered in cases of high risk for recurrence such as dedifferentiated type.

## Conclusions

We encountered a case of dedifferentiated liposarcoma in the spermatic cord. Accurate histopathological diagnosis of liposarcoma was still clinically challenging, but correct pathological diagnosis is necessary for the provision of adequate treatment in order to reduce the risk of recurrence.

## Statement of Ethics

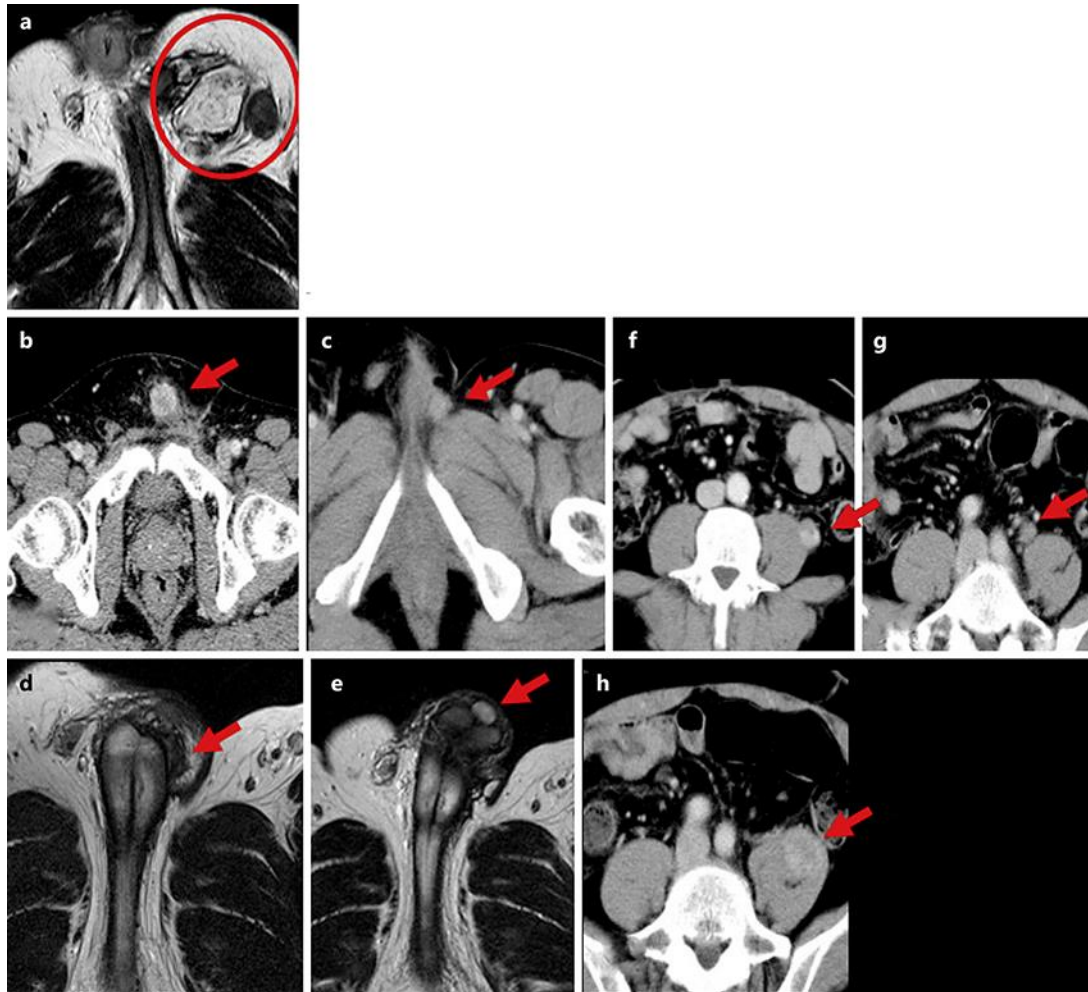
The authors have no ethical conflict to disclose.

## Disclosure Statement

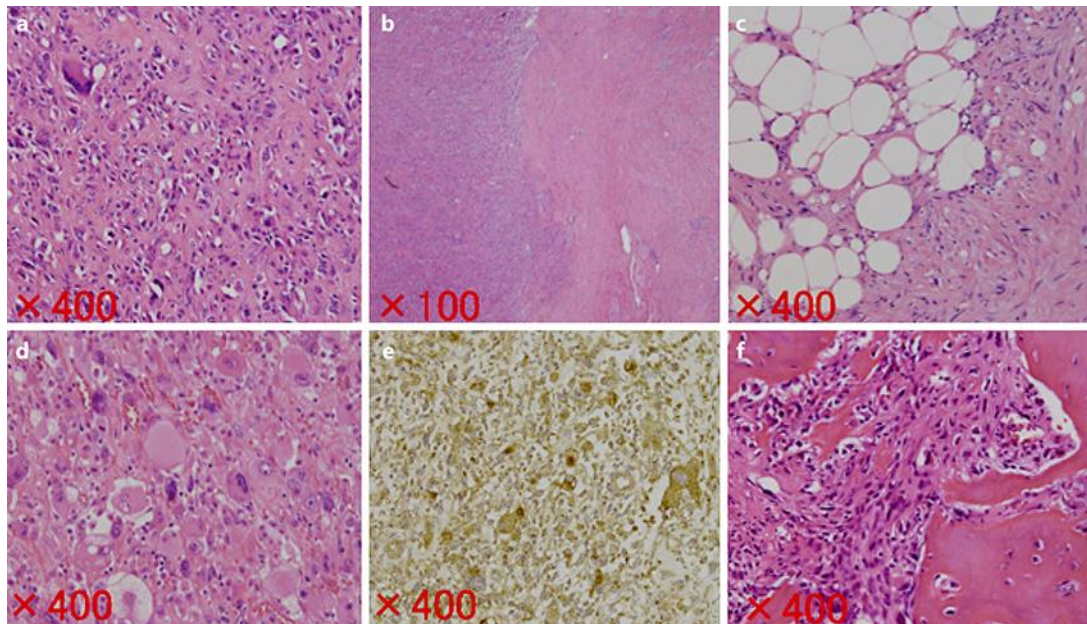
The authors declare no conflict of interest in association with this article.

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**Fig. 1.** Radiographic imaging. **a** CT in July 2006, at first surgery, shows a 30-mm signal hypointense mass in the left spermatic cord. CT and MRI show local recurrence in July 2008 (**b**), retroperitoneal metastasis in January 2009 (**c**), local recurrence in September 2010 (**d**), retroperitoneal metastasis in March 2012 (**e**), local recurrence in September 2012 (**f**), retroperitoneal metastasis in January 2014 (**g**), and local recurrence in May 2014 (**h**).



**Fig. 2.** Histopathological findings. At high orchiectomy in July 2006, staining with hematoxylin and eosin revealed osteosarcoma (a) and well-differentiated liposarcoma including spindle cells and lipoblasts (c), which are characteristic of a biphasic pattern for dedifferentiated liposarcoma (b). e, f Rhabdomyoblastoma with hematoxylin and eosin staining (d) and myoglobin-positive immunostaining (e) at metastasectomy of the psoas muscle in January 2009. f Osteogenesis around the tumor cells at metastasectomy of the psoas muscle in January 2014.

**Table 1.** Histopathological findings in primary and recurrent tumors at every surgery

Operation date	Tumor sites	Operation	Histopathological findings
July 2006	spermatic cord	high orchiectomy	leiomyosarcoma
July 2008	anterior surface of pubic bones	metastasectomy	leiomyosarcoma
January 2009	anterior surface of psoas muscle	metastasectomy	rhabdomyosarcoma
October 2010	penile base	tumorectomy	rhabdomyosarcoma
March 2011	anterior surface of psoas muscle	metastasectomy	rhabdomyosarcoma
September 2012	penile base	tumorectomy	rhabdomyosarcoma
January 2014	intramuscular of psoas muscle	metastasectomy	osteosarcoma

The initial pathological diagnosis was leiomyosarcoma, which was finally diagnosed as dedifferentiated liposarcoma after several recurrences by a thorough overview of histopathological findings of all surgical specimens.