Case Report

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A Giant Paratesticular Liposarcoma

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Abstract

Liposarcoma is a tumour originating from the mesoderm, which captures nearly twenty percent of all sarcomas. It first described by Lesauvage in 1845. Seventy percent of cases are extremity and retroperitoneum masses. Paratesticular liposarcoma is a rare entity. For treatment, a tumour-free margin radical orchiectomy with wide local excision and high ligation of the spermatic cord should be performed. If needed radiotherapy should be applied. An eighty-six-year-old male patient with paratesticular liposarcoma is reported in this article.

Keywords: Orchiectomy, sarcoma, testicular tumor, urologic oncology

Introduction

Liposarcoma is a tumor originating from the mesoderm, which captures nearly twenty percent of all sarcomas. It was first described by Lesauvage in 1845 (1). There are four known histological subtypes; well differentiated, dedifferentiated, myxoid and pleomorphic (1,2). As a soft tissue malignancy, liposarcomas are largely found in areas like head and neck, extremities, gastrointestinal tract and retroperitoneum (3). Seventy percent of cases are extremity and retroperitoneum masses (4). Paratesticular liposarcoma is a rare entity, with about two hundred case reports. Additionally, few these tumors are larger than ten centimeters, referred to as giant liposarcoma (1). It is mostly seen in the elderly population (4). We will present a paratesticular liposarcoma case. Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Case Report

Eighty-six-year-old male patient presented with a swelling in his left scrotum, which had been growing slowly for two years and recently caused pain. On physical examination, it was found that the left scrotum reached a size of at least fifteen centimeters and the ipsilateral testicle inside was not palpable. The patient underwent scrotal Doppler ultrasonography and blood tests were performed. The patient's full blood count values and testicular tumor marker test results were within normal limits. On ultrasonography, a solid mass lesion with an extra testicular location in the left scrotum, associated with the spermatic cord, reaching a diameter of fifteen centimeters, containing locally fatty tissue echoes was observed. To the proximal of this mass, there was a second mass lesion with a similar feature reaching eight centimeters in diameter in the inguinal canal. The total dimension of the two masses reached twenty-five centimeters. To confirm the diagnosis and check for abdominal and thoracic metastases, contrast-enhanced computed tomography (CT) was performed. No other mass was found. The features of the mass were also were consistent with liposarcoma on CT.

In the surgery, the mass was reached by inguinal incision. The adhesions around it have been removed. It was observed that the mass extended to the spermatic cord. Invasion was detected macroscopically with the ipsilateral testis. During orchiectomy, mass removal was performed from the highest level of the spermatic cord that could be detected. After removal, a second specimen was removed from the residual spermatic cord with a new resection to determine the surgical margin. The surgery was completed without complications. The patient was discharged after one day of follow-up.

By the pathological evaluation, the gross examination of the well circumscribed lobulated paratesticular mass, the longest diameter was twenty-one centimeter (Figure 1). Hypo-spermatogenetic testis was entirely rounded by a tumor diagnosed as liposarcoma. Histopathologically, in most areas the liposarcoma was well-differentiated type without necrosis (Figure 2). Nevertheless, there were some areas containing

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prominent myxoid stroma in about 20% of the tumor (Figure 3). However, classical pattern of myxoid liposarcoma was detected in only one slide. Since fluorescence *in situ* hybridization examination for the *MDM2* gene applied for differential diagnosis between myxoid and well-differentiated liposarcoma types showed prominent amplification, the diagnosis of well-differentiated liposarcoma was made. Additionally, in one side tumor was characterized by increased cellularity and minimal atypia, which was interpreted as a possible early dedifferentiation area. The surgical margins were clear.

Discussion

Paratesticular liposarcoma is usually found in men over the age of forty with slowly developing scrotal swelling and stiffness (1,4). Pain is not essential, even if there is, it may develop slowly. They are difficult to distinguish from testicular masses, especially when the mass originates from the spermatic cord and extends to the lower part of the scrotum. It should also be distinguished from diseases such as inguinal hernia, epididymitis, or cord lipoma. Especially, cross-sectional imaging including the abdomen is valuable because it shows the inguinal canal level of the mass, the condition of the retroperitoneal lymph nodes and distant metastases. Biopsy was not recommended for diagnosis. Performing CT or magnetic resonance imaging (MRI) can lead to a more definitive diagnosis. CT and MRI are successful in diagnosing liposarcomas and detecting compression or invasion of nearby organs. Some imaging studies, especially with retroperitoneal liposarcomas, found MRI more successful than CT in diagnosis and follow-up (5). Additionally, they provide high success in determining the histological type and stage of the tumour, with again MRI being better (5,6). Although there is no strong recommendation for the diagnosis, combined positron emission tomography (PET)/CT has been found to be particularly useful in detecting and staging recurrence in the follow-up after surgery or radiotherapy. Combined PET/CT is also superior to using these two methods alone (7).

For treatment, a tumour-free margin radical orchiectomy with wide local excision and high ligation of the spermatic cord is determined as the gold standard (1,4,8,9,10). Recurrence is strongly associated with incomplete excision, and care should be taken to prevent tumor spilling during the procedure (1,4). If margin status is positive or suspicious, radiotherapy should be applied to the inguinal area and to the scrotum depending on the situation (8,9,10). Studies reported that with adjuvant radiotherapy there was no recurrence in median eighteen months of follow-up (11). It should be known that radiation treatment is effective for local control and for positive margin cases. Recurrence was associated with a poor prognosis. The effects of radiotherapy on the other testicle and the adjacent organs should be closely monitored. Data on the efficacy of adjuvant chemotherapy in paratesticular liposarcoma are limited. This is mostly because these masses are rare for sufficient studies and practice. Vincristine, cyclophosphamide and doxorubicin are known suitable agents for metastatic or positive margin paratesticular liposarcomas (12). For the follow-up, particularly cross-sectional imaging is recommended, starting at three months, then at six months, and annually thereafter. It is argued that the total follow-up time should be a minimum of ten years (4).

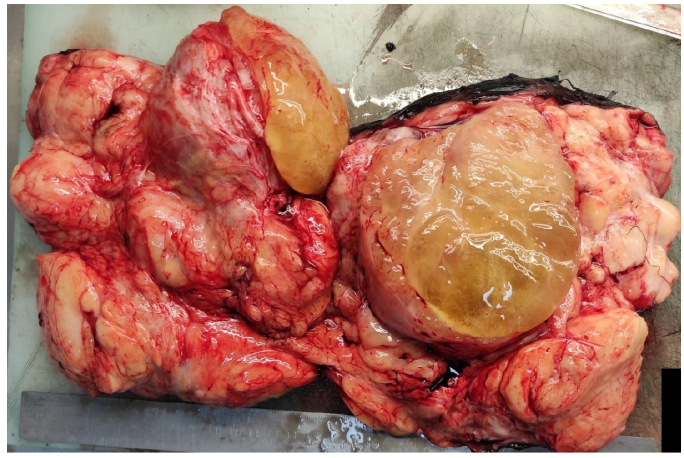

Figure 1. Macroscopically, the paratesticular tumor was lobulated and the cut surface was yellow and fatty

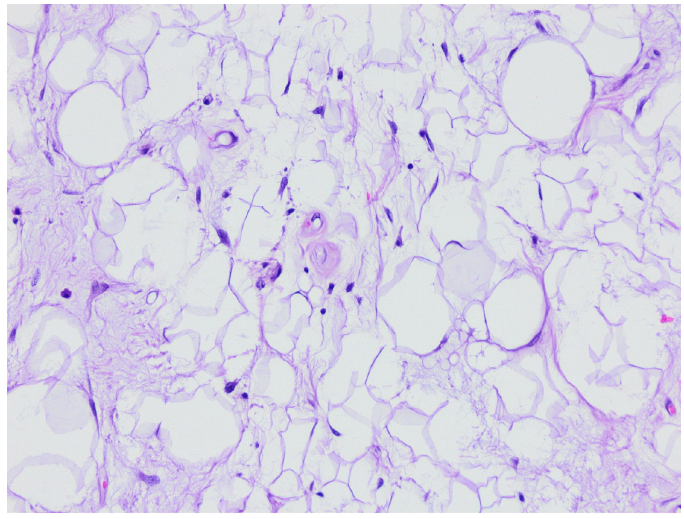

Figure 2. The well-differentiated areas of the tumor (HE, x200)

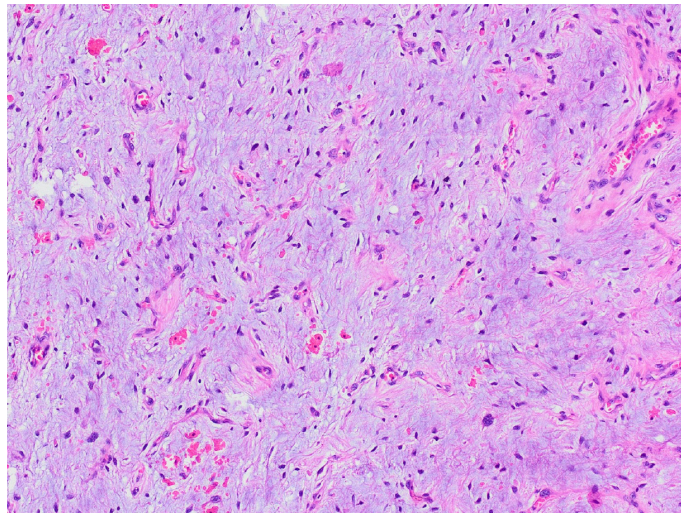

Figure 3. The areas reminiscent myxoid liposarcoma (HE, x200)

In a study of primary retroperitoneal well-differentiated liposarcoma cases that treated surgically, concomitant organ resection was observed in nearly half of the cases. However, the invasion was found to be approximately 15% in the pathology reports (13). Again, in this study, it was argued that routine concomitant organ resection did not have a positive effect on overall survival and disease-free survival, but in contrast, it increased the complication rate and hospital stay. In a similar study investigating the characteristics of salvage surgery in patients with recurrent retroperitoneal well-differentiated liposarcoma, it was observed that routine concomitant organ resection was applied to more than 50% of the patients, but the actual invasion was still about 15% (14). It has also been shown that concomitant organ resection increases the complication rate and hospital stay in these patients too. Both of these single-centre studies with retroperitoneal masses suggest the preservation of uninvolved organs if surgically possible. But as in our case, organ resection with the mass is inevitable in surgically detected invasions. Additionally, it should be remembered that these two studies were conducted with patients with retroperitoneal masses, not paratesticular masses, and because of anatomical features, it is riskier to intervene with nearby structures in the abdomen. In conclusion, it should not be forgotten that high ligation of the spermatic cord with tumour-free margins is the main treatment for paratesticular liposarcoma, as in this study.

Liposarcomas are tumors originating from the mesoderm, which are relatively rare and require a multidisciplinary approach. It should be kept in mind that diagnosis may be delayed due to anatomical features or patient's habits. Providing tumour-free surgical margin and applying suitable radiotherapy if necessary are key points in the treatment. Long-term cross-sectional imaging at regular intervals, including screening for distant metastases are necessary to follow-up.

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Ethics

Informed Consent: Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Design: O.E., Data Collection or Processing: O.E., Analysis or Interpretation: T.M., Literature Review: N.N., Critical Review: N.N., T.M., Writing: O.E.

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