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Case report

Liposarcoma of the Spermatic Cord – A Rare Pathological Entity

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A B S T R A C T

Liposarcoma of the spermatic cord is a very rare neoplasm with fewer than 200 cases recorded in world literature. This report describes a case of liposarcoma of the spermatic cord which developed after radical prostatectomy and salvage radiotherapy for prostate cancer treatment. Four years following surgical treatment of the primary neoplasm and one year following radiotherapy, the 67 –year –old patient was referred to the urology clinic for the emergence of a mass in the right hemiscrotum region. Ultrasonography revealed a homogenous isodense mass measuring 4.5 cm in diameter, while positron emission tomography (PET) with 2-deoxy-2(^{18}F) fluoro-D-glucose (FDG) indicated the presence of viable tumors cells. Radical orchiectomy was performed to remove the neoplasm which appeared to be dedifferentiated liposarcoma of the spermatic cord. Lymph nodes were not affected, but the surgical margins were found positive and the patient was referred to further oncologic treatment.

Key words: *liposarcoma, spermatic cord, prostate cancer, radiotherapy, positron emission tomography*

Introduction

Although most frequently occurring among soft tissue sarcomas, liposarcomas are rarely seen involving spermatic cord structures and are represented with only 3-7 % of all paratesticular sarcomas[1-4]. To the best of our knowledge, it has been only one report describing liposarcoma of the spermatic cord following radical prostatectomy for prostate cancer treatment[5]. Preoperative diagnosis is uncommon, as it is next to impossible to distinguish liposarcoma of the spermatic cord from other pathological entities with similar clinical manifestation, thus an accurate diagnosis can only be obtained by histopathological evaluation of the neoplasm following biopsy and surgery. The recommended treatment is radical orchiectomy with wide local incision while lymphadenectomy is not routinely done since malignancy seldom spreads into the regional lymph nodes. Most liposarcomas of the spermatic cord are well differentiated with low grade of malignancy, so in most cases the prognosis is fair, especially if surgical margins were found clear[6] . However, long-term follow-up is mandatory hence recurrence of the disease is frequently observed[1,2,4,7,8].

Case Report

A 67-year-old man was referred to the urology clinic following the emergence of a mass in the right hemiscrotum region of four month's duration. Four years prior to admission, in August 2008, the patient had undergone retropubic radical prostatectomy (RRP) for the treatment of localized prostate cancer. Preoperative prostate serum specific antigen (PSA) was 9.4 ng/ml and histopathological evaluation classified the neoplasm as adenocarcinoma, Gleason sum 7(3+4), pT2c R1. In January 2009, the PSA level was 0,05ng/ml; however, the biochemical recurrence was noticed in February 2011 when the PSA level reached the value of 0.2ng/ml.

Although computed tomography (CT) of the abdominal and pelvic cavity along with skeletal scintigraphy showed no evidence of the disease recurrence or metastatic process, the patient was submitted to the salvage radiotherapy performed with 66 Gy in 33 fractions 3D CRT due to biochemical evidence of disease recurrence.

Approximately one year following salvage radiotherapy forming of the mass in the right hemiscrotum became noticeable. Ultrasonography revealed a homogenous isodense

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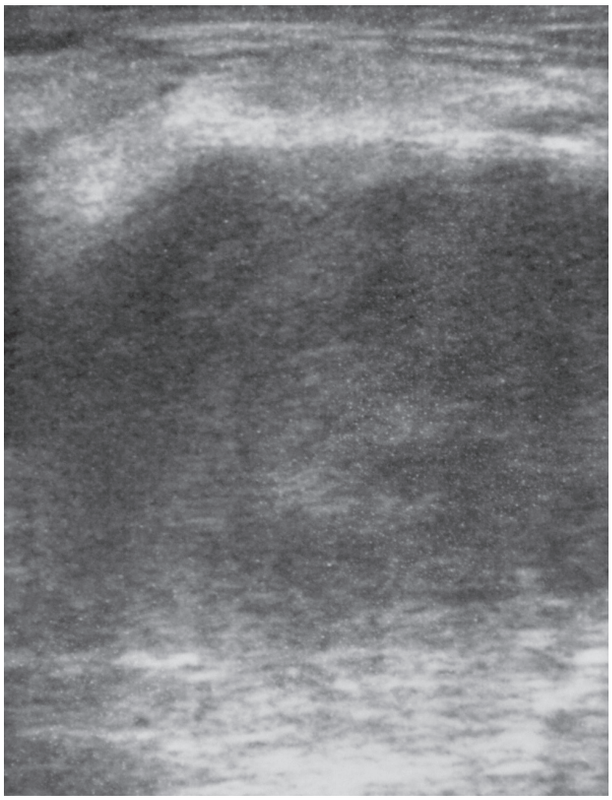

Figure 1. In the right hemiscrotum area ultrasonography revealed a homogenous isodense mass measuring 4.5 cm in its diameter.

mass in the right hemiscrotum area measuring 4.5 cm in diameter (Figure 1). Positron emission tomography (PET) with 2-deoxy-2(^{18}F) fluoro-D-glucose (FDG), which has been used to detect viable tumor tissues on the basis of increased glucose metabolism, showed an intense focal

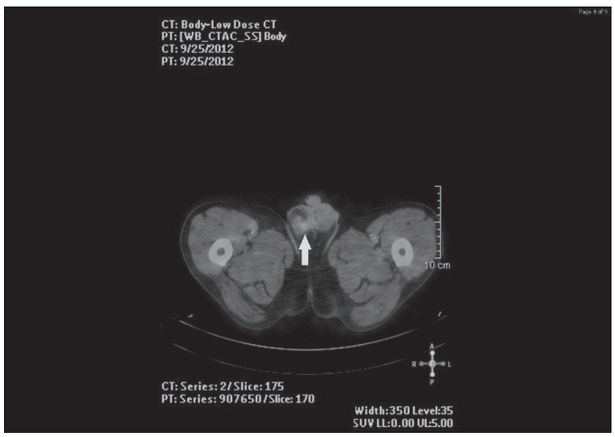

Figure 2. PET scan with FDG radiotracer showed an intense focal FDG uptake in the right hemiscrotal region (green arrow), with no increased FDG accumulation in the regional lymph nodes.

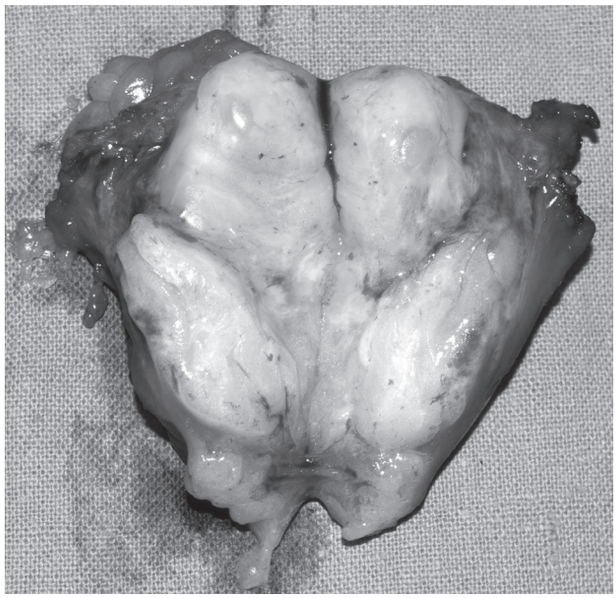

Figure 3. Surgical specimen showing soft encapsulated mass measuring 4.5 cm in diameter that infiltrated right funicular structures.

FDG uptake in the right hemiscrotal region, with no increased FDG accumulation in the regional lymph nodes (Figure 2).

In July of 2012, the radical orchiectomy with wide local excision was performed to remove the neoplasm which appeared to be soft encapsulated mass measuring 4.5 cm in diameter infiltrating right funicular structures (Figure 3). Histopathological examination revealed well-differentiated liposarcoma consisting mainly of lipoblasts with infiltrated dedifferentiated sections composed of atypical round and spindle cells with clusters of large polymorphic multinuclear cells (Figure 4). The tumor was classified as liposarcoma grade 2 according to FNCLCC grading system. Surgical margins were found positive, so the patient was scheduled for further oncologic treatment according to AI protocol x 6. Doxorubicin 40 mg i.v. D1-D3, Holoxan 3000 mg i.v. D1-D2 and 2000 mg D3, Uromitexan 3 x 800 mg i.v. D1-D3 were administered. Approximately one year following chemotherapy multislice computed tomography (MSCT) of the chest, abdomen and pelvis showed no evidence of tumor recurrence or metastatic disease.

Discussion and Conclusion

Considering that the second neoplasm developed a little more than a year following salvage radiotherapy, its association with radiation exposure seems unlikely. It has been estimated that about 8 % of second solid cancers might be related to radiotherapy, but the time frame in which secondary malignancy usually appears is thought to be at least 5 years[9,10]. On the other hand, prostate cancer patients are often diagnosed with multiple malignant neoplasms[11], suggesting that perhaps, in this case, rather

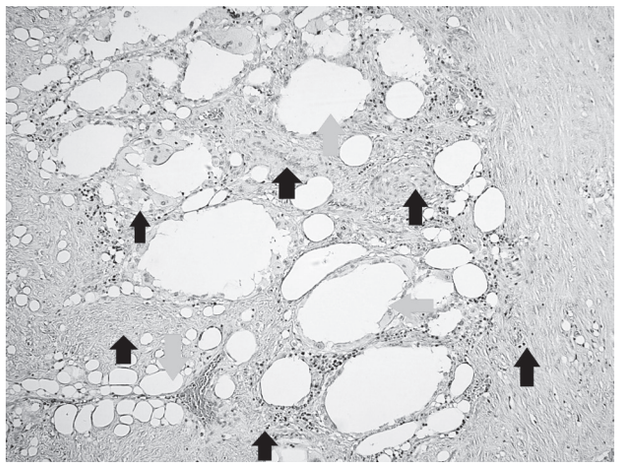

Figure 4. Micrograph showing well-differentiated liposarcoma consisting mainly of lipoblasts (green arrows) with infiltrated dedifferentiated sections composed of atypical round and spindle cells with clusters of large polymorphic multinuclear cells (blue arrows).

than radiation, suppressed function of the immune system had a part in triggering second neoplasm growth.

Liposarcomas usually present as slow growing masses of the inguinal canal or the scrotum, mimicking testicular and epididymal tumors, inguinal hernias, hydrocele, chronic epidydimitis or funicular cysts and are most often suspected intraoperatively and diagnosed postoperatively. An accurate diagnosis can only be obtained by histopathological examination, whereas ultrasonography, CT and MRI scans may serve to visualize the structure and detect lipomatous nature and of the tumor, but without the possibility of obtaining crucial information of tissue microstructure and quality. A great majority of the spermatic cord tumors are benign, among which the lipoma is the most common accounting for nearly 80 % of cases[6]. Malignant tumors of the spermatic cord are most frequently sarcomas; rhabdomyosarcomas mostly develop in younger men and are usually associated with poor prognosis, whereas leiomyosarcomas, fibrosarcomas, and liposarcomas are more frequently seen in older population[5]. Other types of spermatic cord sarcomas are histiocytoma and other histologic subtypes, and all of those have a high propensity for local recurrence[12]. Liposarcoma may arise from fatty tissue in the spermatic cord, or may be a result of uncommonly seen malignant transformation of preexisting lipoma[11]. On CT and MRI scans liposarcoma shows as low density and well-demarcated area[6], while on MRI T2 weighted images it has slightly low signal intensity[5]. PET scan with FDG radiotracer is also not listed among standard diagnostic procedures, but can be of service in recurrent cases and post treatment monitoring. In this way, monitoring of intense incorporation of FDG in tumor tissues can be used to differentiate recurrent tumor from postradiation necrosis, which is otherwise indistinguishable by plain CT and MRI scans.

There are four basic histologic types of liposarcoma: well differentiated, dedifferentiated, myxoid and pleomorphic. Within well-differentiated type of liposarcoma four histologic subtypes have been recognized: adipocytic or lipoma –like, sclerosing, and very rare subtypes of inflammatory and spindle cell liposarcoma[5]. Liposarcomas of the spermatic cord most frequently belong to well-differentiated type with low grade of malignancy and usually do not spread into the regional lymph nodes, but can be locally invasive. Highly malignant types like pleomorphic are rare and readily metastasize into the regional lymph nodes, or even via hematological route to lungs and bones[2,6].

The treatment of choice is radical orchiectomy with high ligation of the spermatic cord and wide local excision[3,4]. In most cases with negative surgical margins the prognosis is excellent; however, local recurrence rate following resection alone has been between 30 and 50%[5], as tumor cells frequently infiltrate adjacent tissues through the pseudo –capsule[6]. Adjuvant radiotherapy is usually not required, except in cases with positive margins and poor prognostic factors[4]. In some cases local recurrence of the disease can be late, even after 20 years, so the usual protocol of 5 years monitoring is not considered adequate to assess the outcome[5,6]. For that reason, the long-term follow-up is mandatory to prevent recurrence and possible increase in grade of malignancy[1-4,7,8].

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LIPOSARKOM SJEMENOVODA –RIJEDAK PATOLOŠKI ENTITET

S A Ž E T A K

Liposarkom sjemenovoda je rijetka novotvorevina sa manje od 200 slučajeva zabilježenih u svjetskoj literaturi. Ovo je prikaz bolesnika sa liposarkomom sjemenovoda koji se pojavio nakon operacijskog zahvata radikalne prostatektomije, a potom izvršene "salvage" terapije zračenjem. Četiri godine nakon kirurškog liječenja primarnog tumora i godinu dana nakon terapije zračenjem, 67 -godišnji pacijent upućen je na urološku kliniku zbog pojave mase u desnoj hemiskrotalnoj regiji. Pregled ultrazvukom otkrio je masu ujednačene gustoće, promjera 4,5cm, dok je pozitronska emisiona tomografija (PET) sa 2-deoksi-2(^{18}F) fluoro-D-glukozom (FDG) upućivala na postojanje metabolički aktivnih tumorskoh stanica. Učinjena je radikalna orhiektomija sa širokom ekscizijom okolnog tkiva, kojom je uklonjen tumor, te je otkriveno da se radi o dediferenciranom liposarkomu sjemenog užeta. Ingvinlni limfni čvorovi nisu bili zahvaćeni tumorskim satnicama, no resekcijski rubovi unatoč širokoj eksciziji tumora su bili pozitivni, te je bolesnik upućen na daljnje onkološko liječenje.