



LIPOSARCOMA OF THE SPERMATIC CORD – DIAGNOSTIC AND THERAPEUTIC ISSUE

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SUMMARY – Liposarcoma of the spermatic cord is a malignant tumor so rare that there are less than 200 cases reported in the literature worldwide. Liposarcoma is a malignancy which originates from fat tissue. Although only 3%-7% of all paratesticular sarcomas primarily arise from structures of the spermatic cord, clinical significance of these tumors must not be neglected because they are often preoperatively misdiagnosed. A 66-year-old male presented with a painless swelling on the left side of the scrotum. Local examination revealed a solid, smooth, limited mass of approximately 4x3 cm in the left side of the scrotum. Tumor markers were within the reference range. Ultrasound examination showed a solid, clearly limited non-homogeneous mass of 40x20 mm localized in the left spermatic cord. Magnetic resonance imaging showed an expansive mass measuring 60x85x60 mm in the left inguinoscrotal region without propagation into the abdominal cavity. Both testicles and epididymides appeared normal on magnetic resonance examination and no locoregional enlarged lymph nodes were seen. The patient was treated operatively with radical inguinal orchiectomy. In conclusion, liposarcomas of the spermatic cord are extremely rare neoplasms that clinically present as slow-growing, painless, palpable inguinal or scrotal masses. Radical orchiectomy with high ligation of the spermatic cord and wide excision of the surrounding soft tissues within the inguinal canal remains the gold standard treatment option. Recurrence of the disease is frequent even several years after primary therapy, therefore long-term follow-up is mandatory.

Key words: Liposarcoma; Spermatic cord; Magnetic resonance imaging; Radical orchiectomy; Recurrence

Introduction

Liposarcoma is a malignant tumor that originates from mesenchymal cells and represents the most common type of primary malignant tumor of the spermatic cord¹. Paratesticular liposarcomas are very rare conditions and there are less than 200 cases reported in the literature to date. Most of these tumors manifest as painless, slow-growing scrotal or inguinal masses. Given their rare occurrence, spermatic cord liposarcomas are often misdiagnosed as inguinoscrotal hernias,

hydrocele, lipomas, funicular cysts or testicular tumors². Regarding the rarity of these malignancies, there is little information on liposarcomas and no official recommendations and guidelines for diagnosis, treatment and follow-up of these patients. These limitations have led to the lack of consensus on optimal surgical and adjuvant treatment^{3,4}.

Case Report

A 66-year-old male presented with a painless scrotal mass on the left side. The patient reported that he had noticed painless scrotal mass about 6 months before, growing progressively ever since. Physical examination revealed a solid, smooth-surfaced mass measuring 4x3 cm on the left side of the scrotum. Tumor

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markers (human chorionic gonadotropin, alpha-feto-protein and lactate dehydrogenase) were within the reference range. Scrotal ultrasonography revealed an oval, clearly limited, inhomogeneous mass of 40x20 mm within the left spermatic cord. Testicle and epididymis showed no pathologic abnormalities. Magnetic resonance imaging (MRI) showed an expansive mass measuring 60x85x60 mm in the left inguinoscrotal region, which was T2W hypodense and T1W hyperdense. There were no MRI signs of mass propagation to the abdominal cavity. Testicle and epididymis on both sides appeared normal on MRI examination. No enlarged locoregional lymph nodes were found.



Fig. 1. Magnetic resonance image of the scrotum showing liposarcoma of the left spermatic cord.

Based on clinical and radiological findings, surgical exploration of the spermatic cord and scrotum was performed using inguinal approach. The left spermatic cord was dissected and a yellow, multilobulated mass measuring 9x7 cm was found.

The mass involved the entire circumference of the spermatic cord and was descending into the scrotum, surrounding the testicle. Testicle and epididymis appeared normal with no signs of tumor infiltration. Based on MRI and intraoperative findings, it was decided to perform radical orchiectomy with high ligation of the spermatic cord. Postoperative course was uneventful and the patient was discharged on postoperative day 4. Histopathologic finding showed well-

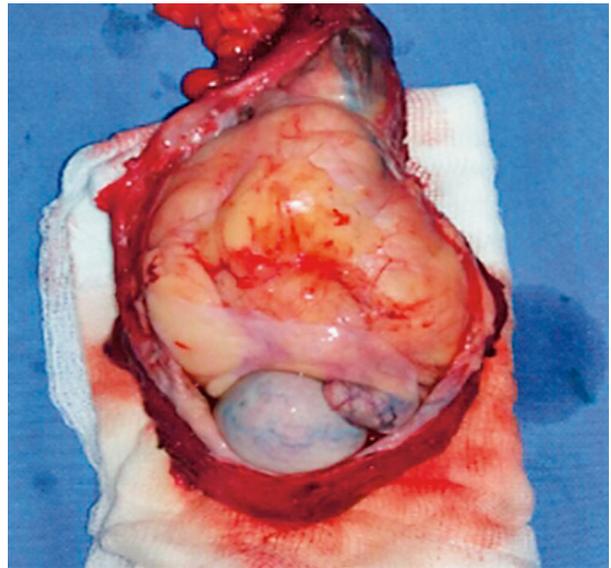


Fig. 2. Intraoperative finding showing bivalved spermatic cord liposarcoma.

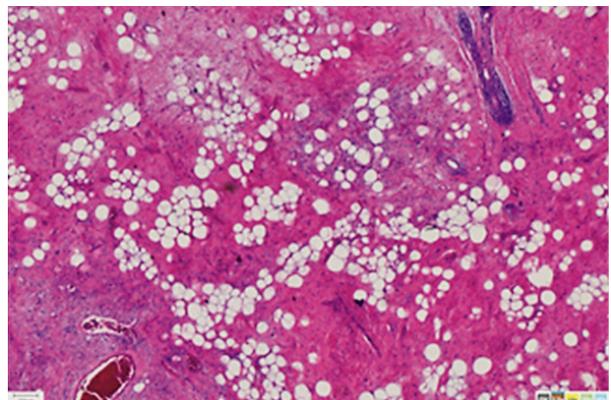


Fig. 3. Well-differentiated (lipoma-like) liposarcoma with sclerosing areas and numerous lipoblasts (HE, X200).

differentiated liposarcoma of the spermatic cord and adenomatous hyperplasia of rete testis.

Six months following the surgery, MRI showed no signs of tumor recurrence and enlarged pelvic lymph nodes.

The patient provided informed consent for the information presented here to be shared.

Discussion

In the adult population, more than 75% of primary paratesticular tumors arise from the spermatic cord.

Most authors agree that sarcomas primarily originate from mesodermal tissue rather than due to malignant transformation of benign lipomatous tissue⁵. Primary spermatic cord malignancies usually originate below the external inguinal ring and therefore present more often as scrotal masses than inguinal masses. Difficulties in diagnosing these tumors are due to the fact that more frequent conditions manifest themselves as painless scrotal masses. Therefore, spermatic cord liposarcomas are often misdiagnosed as inguinoscrotal hernias, lipomas, hydrocele, epididymal cysts, or testicular tumors^{6,7}. Most spermatic cord liposarcomas are well differentiated, low-grade malignancies with no or minimal tendency to metastasize. Liposarcomas are locally aggressive tumors with a high incidence of local recurrence⁸.

Considering the numerous pathologic conditions in the differential diagnosis of scrotal masses, first-line examination should be ultrasound (US). The usual US presentation of spermatic cord liposarcoma is a heterogeneous and hypervascular mass with hyperechoic areas that represent different amounts of fat inside the tumor^{9,10}. Although US can provide useful information regarding location, size and consistency of the mass, there are no pathognomonic sonographic features that can differentiate benign from malignant lesions, so additional imaging is often necessary^{11,12}.

Computerized tomography (CT) and MRI are superior imaging diagnostic tools which can be more helpful in assessing the exact size, location and consistency of tumor, as well as evaluating the status of spermatic cord and testicle. According to some authors, these diagnostic methods may be helpful in narrowing the differential diagnosis by suggesting fat-containing tumors such as liposarcomas¹³. Definite diagnosis is usually made after histopathologic examination, considering that primary spermatic cord tumors do not show specific radiographic signs or patterns⁴.

General recommendations for treatment are based on case reports and small series due to the rare occurrence of these tumors⁴. Radical orchiectomy with high ligation of the cord and wide tumor resection with no microscopic positive margins is the essential element in surgical management of primary spermatic cord malignancies^{13,14}.

Some authors consider that local recurrence of these tumors is more likely a consequence of incomplete excision rather than differentiation level, histo-

logic type and tumor size^{15,16}. In addition, the tendency of sarcomas to infiltrate local structures significantly increases the difficulty to complete radical resection, and positive margins can actually promote tumor seeding through the operative site¹⁷.

A recent publication by Coleman *et al.*¹⁵ proposes extensive *en bloc* excisions of all surrounding tissues that are potentially infiltrated and re-excision in case of local recurrence. They have reported that nearly one-third of the patients without clinically manifest disease who underwent re-excision were found to have residual tumor. A decrease in the local recurrence rate and improvement in disease-free survival was shown in patients having undergone reoperative wide excision after prior incomplete resection¹⁵. Other authors showed long disease-free periods after simple tumor excision without radical surgical approach such as radical orchiectomy¹⁸. Regardless of the above, radical orchiectomy with high spermatic cord ligation is considered the gold standard treatment for spermatic cord liposarcomas according to most authors.

The role of adjuvant radiation therapy and chemotherapy for treatment of spermatic cord sarcomas has not been well established^{4,15}. However, due to the high risk of local recurrence following surgery, there is increasing evidence that all spermatic cord tumors regardless of histologic type and grade should receive adjuvant radiotherapy. Therefore, the most suitable treatment option might be a multimodality approach including radical orchiectomy and adjuvant radiotherapy. Spermatic cord sarcomas have shown resistance to chemotherapy, so routine systematic chemotherapy is not recommended^{19,20}. In addition to all the above, it is important to point out that there is an increasing incidence of testicular cancer in our country, as well as in neighboring countries²¹.

Conclusion

Liposarcomas of the spermatic cord are very rare malignant tumors, and therefore they are often preoperatively misdiagnosed. Liposarcomas present locally aggressive malignancies and show a high incidence of local recurrence. There is no general consensus regarding proper treatment and management of the disease. Radical orchiectomy with high ligation of the cord and wide tumor resection with no microscopic positive margins is the most recommended treatment option

according to most authors. The role of adjuvant radiation therapy and chemotherapy in the treatment of spermatic cord sarcomas still remains controversial. Long-term patient follow-up is mandatory because local recurrence of the spermatic cord liposarcomas is common even several years after primary therapy.

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Sažetak

LIPOSARKOM SJEMENSKE VRPCE – DIJAGNOSTIČKI I TERAPIJSKI IZAZOV

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Liposarkom sjemenske vrpce je toliko rijedak maligni tumor da je do sada opisano manje od 200 slučajeva u literaturi. Liposarkom je malignitet koji vodi podrijetlo od masnog tkiva. Iako samo 3%-7% svih paratestikularnih sarkoma primarno potječe od struktura sjemenske vrpce, kliničko značenje ovih tumora ne smije se zanemariti, jer ih se često prijeoperacijski pogrešno dijagnosticira. U radu je prikazan bolesnik u dobi od 66 godina s kliničkom slikom bezbolne tumefakcije hemiskrotuma s lijeve strane. Lokalni nalaz je ukazivao na solidnu, glatku, jasno ograničenu masu veličine oko 4x3 cm u lijevom hemiskrotumu. Vrijednosti tumorskih biljega bile su unutar referentnih vrijednosti. Na ultrazvučnom pregledu opisana je solidna, jasno ograničena nehomogena masa dimenzija 40x20 mm u lijevoj sjemennoj vrpci. Magnetska rezonancija abdomena i zdjelice pokazala je ekspanzivnu masu u lijevoj ingvinskrotalnoj regiji veličine oko 60x85x60 mm bez znakova propagacije u abdominalnu šupljinu. Na magnetskoj rezonanciji testisi i epididimisi su imali normalan izgled i nisu uočeni uvećani loko-regionalni limfni čvorovi. Bolesnik je tretiran operativno, napravljena je radikalna orhiektomija ingvinalnim pristupom. U zaključku, liposarkomi sjemenske vrpce su ekstremno rijetke neoplazme koje se klinički manifestiraju kao sporo rastuće, bezbolne, palpabilne ingvinalne ili skrotalne mase. Radikalna orhiektomija s visokim podvezivanjem sjemenske vrpce i širokom ekscizijom okolnih mekih tkiva unutar ingvinalnog kanala predstavlja zlatni standard liječenja. Recidiv bolesti je čest, čak i nekoliko godina nakon primarnog liječenja te je dugačko razdoblje praćenja obvezatno.

Ključne riječi: *Liposarkom; Sjemenja vrpca; Magnetska rezonancija; Radikalna orhiektomija; Recidiv*