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Liposarcoma of spermatic cord mimicking an inguinal hernia

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Liposarcoma of spermatic cord mimicking an inguinal hernia. A case report and review of the literature.

AIM: *Liposarcoma of the spermatic cord (LSC) is a tumour often mistaken for common inguinal swelling as hernia and the aim of this work is to present our case with a review of the Literature to define the management of this rare condition.*
MATERIAL OF STUDY: *A systematic review has been realised, considering English language articles published on Pubmed, between 1956 and 2022, using as key words "Liposarcoma of the spermatic cord".*
RESULTS: *160 studies described 420 cases of LSC and in 40 cases the patient had undergone surgery with an initial diagnosis of inguinal hernia.*
DISCUSSION: *LSC is a very rare entity of genitourinary malignancies, occurring more often in the spermatic cord and diagnosis can be difficult. Our case and Literature data confirm the role of imaging in not conventional inguinal swelling, to avoid diagnostic mistakes and to define preoperatively the correct surgical management.*
CONCLUSIONS: *Imaging is mandatory in case of diagnostic doubt. The recommended treatment is a radical high orchiectomy with clear margins. A long follow-up period is necessary to detect a local recurrence which may occur even several years after the primary therapy.*

KEY WORDS: Inguinal swelling, Liposarcoma, Spermatic cord

Introduction

Liposarcoma of the spermatic cord (LSC) is a rare tumor of paratesticular tissues accounting for less than 12% of all liposarcomas. Only about 590 cases were described in the literature from 1956 to 2022. Due to the rarity of LSC, the very few cases present in the Literature, and to the broad spectrum of diseases presenting in the inguinal region, the diagnosis and treatment are very challenging for clinicians. We report the case of a well-differentiated liposarcoma of right spermatic cord mimicking an inguinal hernia.

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Materials and Method

We made a systematic review of articles published on Pubmed only in English language. Between 1956 and 2022, using as key words "Liposarcoma of the spermatic cord". We included in our study all clinical cases about LSC which were preoperatively diagnosed as an inguinal hernia.

Results

We found 289 articles and two review articles about LSC. We excluded 129 articles as they were either not published in English or they were not appropriately focused on the clinical subject. One hundred and sixty studies described 420 cases of LSC and in 40 cases the patient had undergone surgery with an initial diagnosis of inguinal hernia (Fig. 1).

Clinical Case

SG, a 44 y.o. male, comes to our outpatient unit for a painless scrotal mass, which was detected after a domestic scrotal trauma occurred 8 years before. An increase in size affecting the right hemiscrotum was detected following an intense physical exercise. Past medical history was negative for any comorbidity.

Before our clinical evaluation, our patient was examined by a urologist who excluded the presence of a right hydrocele and any testicular-epididymal lesions. the mass was referred to be a right inguinoscrotal hernia.

Physical examination revealed a 10 cm-hard-elastic mass occupying the entire right hemiscrotum, non reducible, with no involvement of the external inguinal ring. Even with a Valsalva manoeuvre the inguinal ring appeared unrelated to such lesion.

Inguinoscrotal US showed a voluminous solid mass, dishomogeneously hypoechoic, with some hyperechoic areas. Some features of vascularization were present. It was lodged in the right hemiscrotum and partially in the homolateral inguinal ring. The mass had well defined margins and was separated from the didymus, which showed regular vascularization but was slightly compressed in periphery. There was no ultra-sonographic evidence of hydrocele.

At the abdominal MRI a right voluminous mass was evident, with partial involvement of the homolateral inguinal canal. It was separated from the epididymus, which showed regular vascularization and was compressed peripherally by the lesion. It is composed by solid components with dishomogeneous intensities with some areas of adipose tissue. Scarce and faded contrast medium enhancement was evident. No hydrocele nor intestinal herniation were present. No contralateral inguinoscrotal alterations were detected. The surgical procedure was performed by a multidisciplinary team composed by a general surgeon and a urologist. The aspect was of a yellowish, multilobulated, 15-cm scrotal tumor. The lesion was capsulated, with regular margins, its consistency was lipomatous in periphery and hard in the middle. It was softly adherent to the right testicle and to the spermatic cord without a sure macroscopic infiltration. In consideration of the age of the patient, the tumour's clear margins, the presence of a capsular structure and the absence of a local invasion, showed by the imaging and confirmed during surgery, a "en bloc" resection of the lesion was performed sparing the testicle and the spermatic cord.

At the histological studies an adipocytic neoplasia constituted by atypical spindle-like cells, plurinucleated was found (Fig. 2). These neoplastic cells were disposed mainly in single elements on top of abundant mixo-fibro-hyalinated stroma and occasional lipoblasts. There was

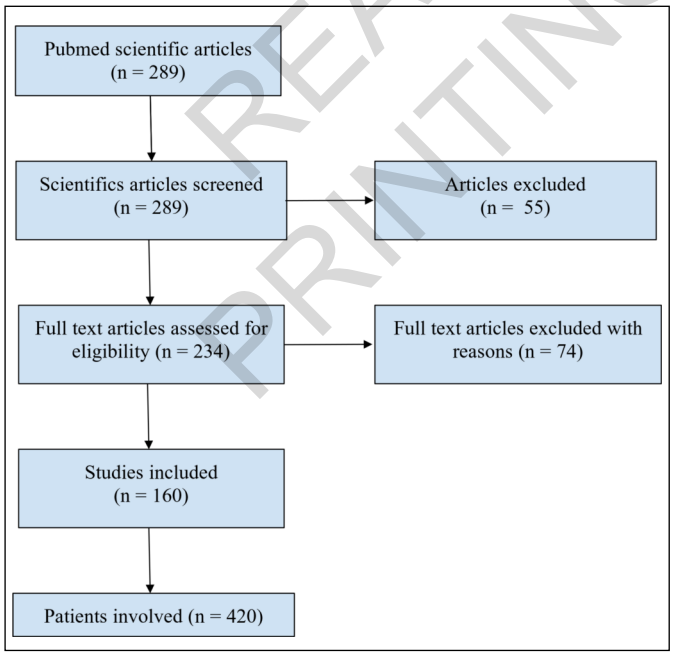

Fig. 1: Flowchart of review of the literature.

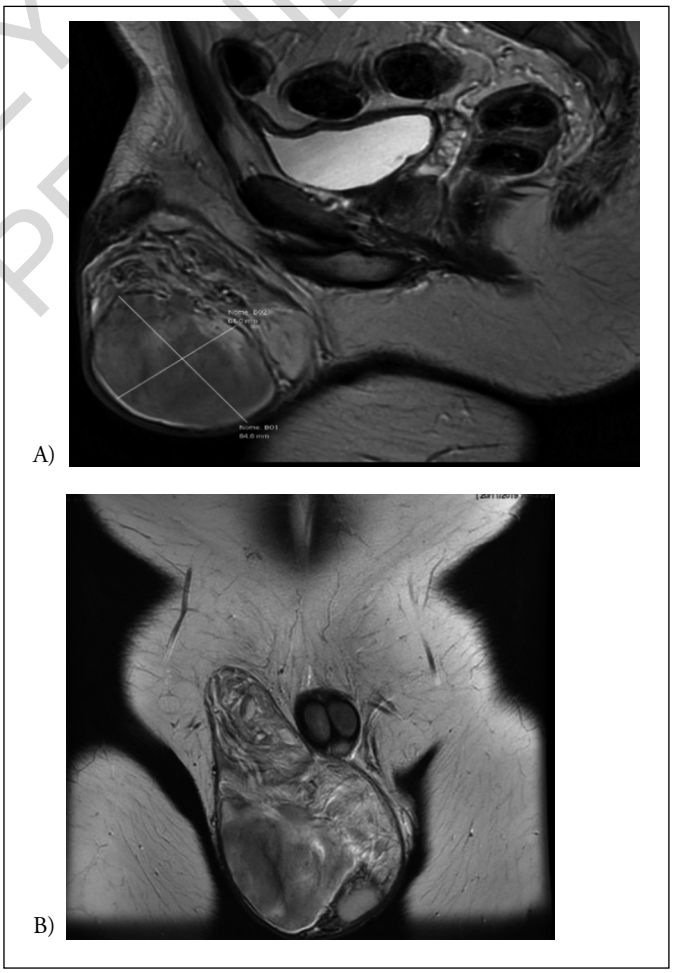

Fig. 2: A) T2 weighted Coronal plane imaging show the complex mass in the inguinal canal. B) T2 weighted Sagittal plane imaging show solid mass between calipers. Above it is the compressed spermatic cord (20x25mm).

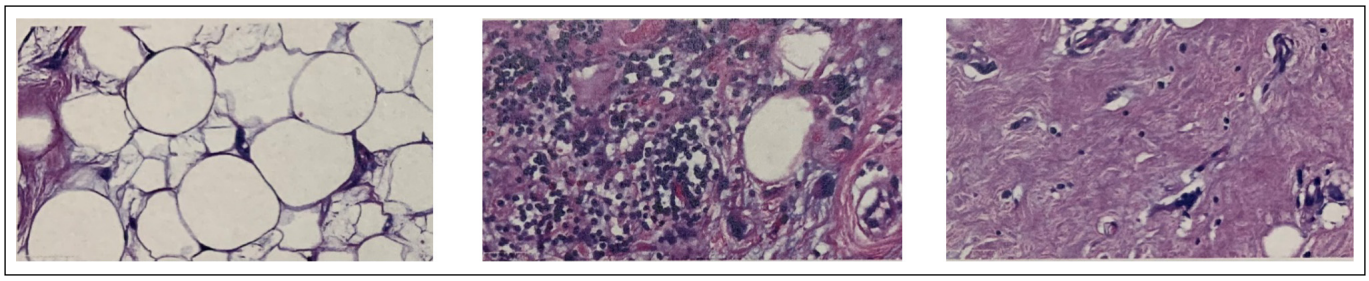

Fig. 3: Histophatologic examination revelead a well differentiated liposarcoma characterized by atypical spindle-like cells, plurinucleated with abundant mixo-fibrohyalinated stroma and occasional lipoblasts.

no vascular nor perineural invasion. The morphological features are suggestive of a well differentiated liposarcoma, sclerosing variant. Margins were widely clean confirming a radical excision (R0).
In consideration of the risk of recurrence and the sparing testicular surgery, after multidisciplinary team meeting, an adjuvant radiotherapy was proposed, later refused by the patient.
No recurrence was detected at 3 years follow-up, after the patient was lost to follow-up.

Discussion

LSC was first reported by Lesauvage in 1845 [1]. Based on the case reports, it appears that the majority of the patients with LSC present in the fifth or sixth decade of life with a painless, irregular, slow-growing inguinal or inguinoscrotal mass clearly distinct from the testis.
LSC is rare variant of genitourinary malignancies, occurring more often in the spermatic cord where it accounts for less than 12% of all liposarcomas [5].
Liposarcomas are the most common soft tissue sarcomas. There are five histological subtypes in liposarcoma, which include well differentiated/atypical lipomatous tumour, dedifferentiated, myxoid and pleomorphic according to the 2013 World Health Organisation (WHO) classification of tumours of soft tissue and bone [8].
Diagnosis of LSC preoperatively may be challenging, as their clinical presentation may be overlapping with other common conditions, including inguinal hernia, lipoma, hydrocele, epididymal cyst, funicular cyst or testicular tumour [9-12]. The typical clinical manifestation of LSC is a slowly growing, non-tender, painless, nodular mass of variable size, located intra-scrotally above the testis or in the groin. Only a few cases are presented with a painful node [11,13,14]
However, the incidence of liposarcomas diagnosed during hernia surgery is very low (less than 0.1%) [18], and lipomatous lesions greater than 10 cm detected during a surgical procedure for hernia should routinely sent for histologic examination [17].
From the 1956 to 2022, 35 cases of LSC with an initial diagnosis of inguinal hernia were described (Table I)
The diagnosis of LSC is based upon radiological investigations, such as Ultrasonography (US), Computerized Tomography (CT) and Magnetic Resonance Imaging (MRI).
The recommended treatment for LSC is a radical high orchiectomy. However, the anatomical features of the inguinal region make it difficult to perform this procedure, and negative resection margins are sometimes close to the tumours. A second resection is advised if the margins are positive, to avoid misleading conclusions [2-4].
Retroperitoneal lymph node dissection should be limited to patients with radiologically suspicious lymph nodes only.
Khandekar et al. found that the 3-year local recurrence free survival was 100% for negative margins compared with 29% for positive margins. Incomplete excision is associated with frequent recurrence, underlying the importance of an aggressive surgical management in order to improve disease-free survival [19]. The prognosis of LSC depends on the histological cell type. The well-differentiated type has a better prognosis [2].
Indications for adjuvant radiotherapy include positive margins or less than 10 mm and when the tumour is not completely resectable, because in these cases local recurrence after surgery alone is very high [16-18]. The adjuvant radiation therapy improved local control but do not seems assess its effect on long-term survival.
The role of adjuvant systemic chemotherapy in adult with LSC is still debated. LSC is resistant to chemotherapy, so this treatment is not routinely recommended., A treatment with vincristine, cyclophosphamide and doxorubicin can be proposed for high grade or/and metastatic LSC [18].
Our case report reinforces the importance of an accurate preoperative imaging. It allowed to confirm the diagnosis and also to perform a tailored surgical strategy with a testicular sparing procedure in a first time, postponing an eventual second look for larger resections on the basis of histological findings.

Conclusion

LSC is a very rare entity. Preoperatively the diagnosis can be difficult and imaging is mandatory in case of diagnostic doubt. All cases of LSC should be discussed

Table I - *Overview of published cases of liposarcoma of the spermatic cord with an initial diagnosis of inguinal hernia.*

Author	Patient (n)	Diagnosis	Histology	Year	Measure	Treatment
Roslyn JJ.	1	Hernia	Liposarcoma	1980		
Torosian MH.	1	Hernia	Low grade liposarcoma	1987	15x20 cm	Total excision
Certo LM.	1	Hernia	Myxoid liposarcoma	1988	5.5x3.5cm	Total excision
Kitamura K.	1	Hernia/lipoma	Well differentiated liposarcoma	1996	15x11x4.5cm	Total excision
Bulbul MA.	1	Hernia	Dedifferentiated liposarcoma	1996	10x7 cm	Radical orchiectomy and en bloc resection of the mass
Montgomery E.	2	Hernia	Well differentiated liposarcoma	1999	13 cm; 10 cm	Total excision
Noguchi H.	1	Hernia	Well differentiated liposarcoma	2001	14x3.8 cm	Total excision
Bouropoulos C.	1	Hernia	Well differentiated liposarcoma	2001	7.5x4.5x4 cm	Radical orchiectomy and en bloc resection of the mass
Panagis A.	1	Hernia	Myxoid liposarcoma	2003	5 cm	Hernia repairing and total excision
Hassan JM.	1	Hernia		2003		Radical orchiectomy
Kalyvas KD.	1	Hernia	Well differentiated liposarcoma	2004	10x9x4 cm	Total excision
Vázquez-Lavista LG.	2	Hernia	Myxoid/pleomorphic liposarcoma	2006	20x15 cm; 20x20 cm	Total excision; hernia repairing and total excision
Baldassarre E.	1	Hernia	Well differentiated liposarcoma	2007	5x4x3 cm	Partially removed
Manzia TM.	1	Hernia	Well differentiated liposarcoma	2010		Total excision
Hsu Y-F.	1	Hernia	Myxoid liposarcoma	2012	6x5x3 cm	Total excision
McKinley SK.	1	Hernia/liposarcoma	Well differentiated liposarcoma	2013	10.3 x7.4 x18.1 cm	Total excision
Li F.	1	Hernia	Well differentiated liposarcoma	2013	6x5x3 cm	Radical orchiectomy and en bloc resection of the mass
Bhullar JS.	1	Hernia	Well differentiated	2013	3x2 cm	Hernia repairing and total excision
Londeree W.	1	Hernia	Well differentiated liposarcoma	2014	14x8x4 cm	Radical orchiectomy
Abolhasani M.	3	Hernia	Well differentiated liposarcoma	2014	10x10x5 cm; 9x7x6 cm; 16.5x8.5x6 cm	Radical orchiectomy
Raza M.	1	Hernia	Myxoid liposarcoma	2014	20x14x5 cm; 16x14x4 cm	Bilateral orchiectomy
Pănuş A.	1	Hernia	Well differentiated liposarcoma	2015	7/8 cm	Radical orchiectomy and en bloc resection of the mass
Valeshabad AK.	1	Hernia	High/low grade liposarcoma	2016		Total excision
Chalouhy C.	1	Hernia	Well differentiated liposarcoma	2017	7x3 cm	Radical orchiectomy and en bloc resection of the mass
Thomas KL.	1	Hernia/liposarcoma	Dedifferentiated liposarcoma	2018	15 cm	Radical orchiectomy and en bloc resection of the mass
Febres-Aldana CA.	2	Hernia	Well differentiated liposarcoma	2018	11x5.5 cm; 14x10.5 cm	Radical orchiectomy; total excision
Mouden K.	1	Hernia	Well differentiated liposarcoma	2019	15x7x17 cm	Total excision
Xenaki S.	1	Hernia	Well differentiated	2019		Hernia reparing and excision
Wetzel E.	1	Hernia	Dedifferentiated liposarcoma	2020	4x4x2.8 cm	Hernia reparing and excision
Dunev VR.	1	Hernia	Well differentiated liposarcoma	2020	2x1.5 cm	Radical orchiectomy and en bloc resection of the mass
Shaban Y.	1	Hernia	Dedifferentiated liposarcoma	2020	9x6x5 cm	Radical orchiectomy and en bloc resection of the mass
Miano ST.	1	Hernia+mass	Well differentiated liposarcoma	2020	2x1 cm	Total excision
Pikramenos K.	1	Hernia	Well differentiated liposarcoma	2022	3.21x2.27 cm	Radical orchiectomy and en bloc resection of the mass
Chan K.	1	Hernia	Well differentiated and dedifferentiated liposarcoma	2022	17x15x8 cm	Radical orchiectomy and en bloc resection of the mass
Pavone G.	1	Hernia	Well differentiated liposarcoma	2022	10 cm	Total excision

by a multidisciplinary team. The recommended treatment is a radical high orchiectomy with clear margins. A testicular sparing surgery could be considered in some cases after an accurate preoperative imaging. A long follow-up period is necessary to detect a local recurrence which may occur even several years after the primary therapy.

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