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Spermatic cord liposarcoma presented as scrotal swelling: a case report

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Abstract

Background Liposarcoma is considered the most common among spermatic cord sarcomas. It usually presents as a progressively enlarging inguinoscrotal mass. The resemblance between the spermatic cord liposarcoma and the inguinal hernia allows the swelling to be tolerated for a long duration giving time for hematogenous dissemination. In this article, we share the experience of a unique case of metastatic liposarcoma of the spermatic cord that showed a stationary course of the lower abdominal satellite lesions on follow-up following palliative resection.

Case presentation We present a case of a 53-year-old male with progressively enlarging painless scrotal swelling over 1-year duration. A heterogeneous lesion of the right hemiscrotum and spermatic cord with non-specific radiological features was described by imaging. Two lower abdominal skip lesions were also discovered synchronously in addition to the primary one and were designated inoperable as they were adherent to the nearby bowel loops. The patient underwent cytoreductive surgery of the primary tumour which was proven to be an undifferentiated liposarcoma with free safety margins based on histopathology. Finally, the patient was referred for adjuvant chemotherapy and sequential follow-up.

Conclusions Despite being a rare tumour, the radiologist should always consider the liposarcoma of the spermatic cord in the differential diagnosis of inguinoscrotal lesions even in the absence of typical imaging features. The diagnosis and management of spermatic cord liposarcoma remain a challenge with no clear accepted guidelines.

Keywords Cancer, Imaging, Liposarcoma, Spermatic cord

1 Background

The inguinal canal in males contains the vas deferens, testicular artery and vein, lymphatic vessels, and nerves which compose the spermatic cord (SC) [1]. Although the pathologic conditions affecting the SC are relatively common in clinical practice [2, 3], malignant lesions of the SC on the other hand are extremely rare [4], with the liposarcoma of the spermatic cord (LSC) being the most prevalent accounting for 3–7% of SC neoplasms [5–9] and usually presents between the 5th and 6th decades of life [10]. The typical presentation of LSC is a progressively enlarging painless scrotal mass [11]. As this presentation is shared by a wide variety of lesions, it is challenging to make a diagnosis based on clinical examination alone [1, 8].

As the SC is the seat of a wide variety of lesions from different entities, the radiologist must become familiar with the characteristic imaging findings in each type [2, 3]. The distinguishing imaging features of each pathology affecting the SC are summarized in Fig. 1. Being readily available, cheap and devoid of radiation hazards, ultrasound (US) is considered the primary imaging modality for inguinoscrotal swellings. Although it provides valuable information, it offers no definite

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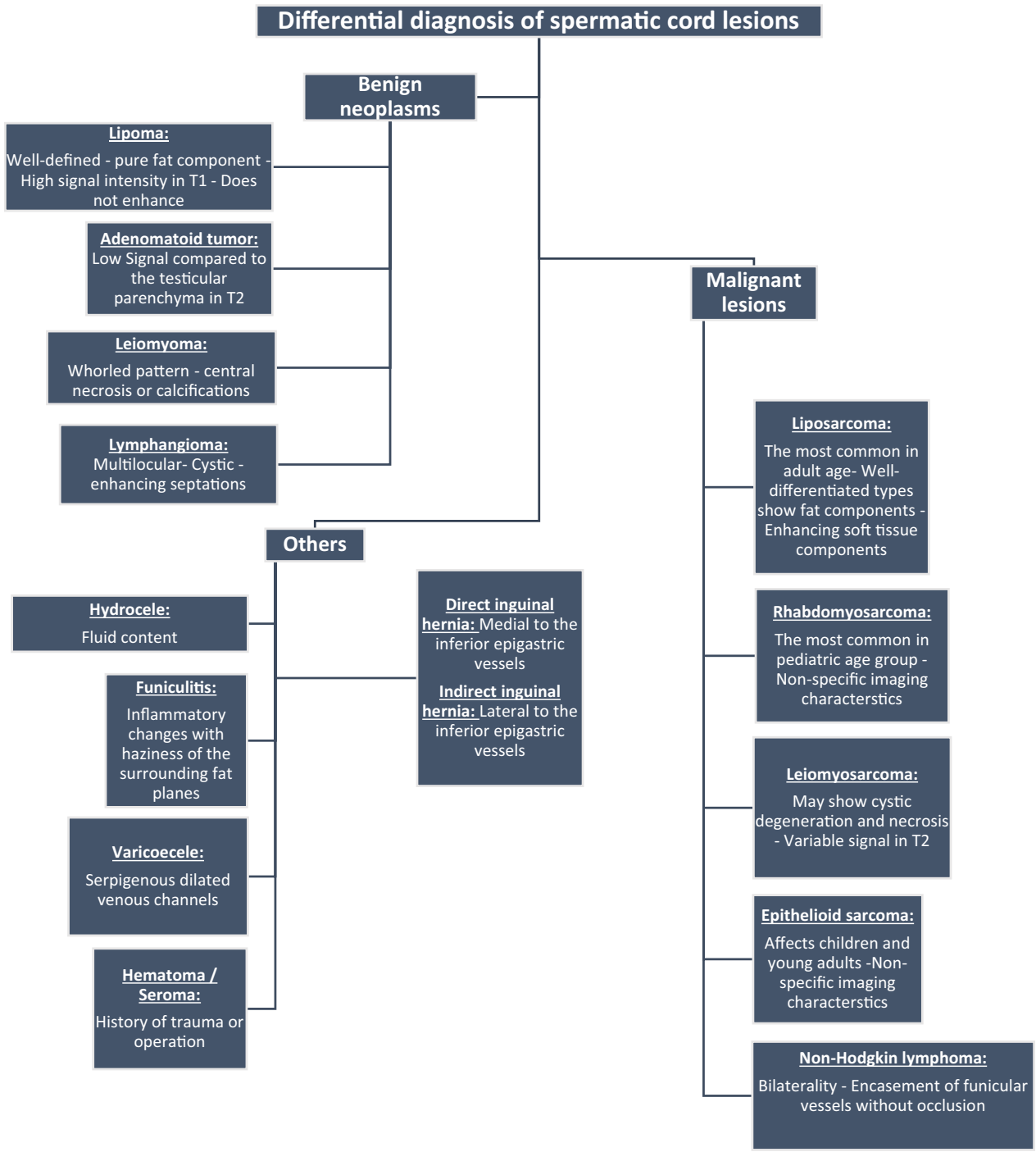

Fig. 1 Differential diagnosis of spermatic cord lesions

discrimination between benign and malignant lesions [7, 12]. Contrast-enhanced computerized tomography (CECT) is usually suggestive of the diagnosis and helps determine the involvement of the anterior abdominal wall or the retroperitoneum [9]. By CECT, LSC usually appears as a heterogeneously enhancing inguinal soft tissue mass with variable areas of fat attenuation [5, 6]. Magnetic resonance imaging (MRI) allows accurate delineation of the tumour margins and tissue characterization [13]. The appearance of LSC by MRI is variable depending on the degree of differentiation [14].

Radical orchiectomy with wide local tissue resection is the accepted surgical treatment for LSC [1, 8]. The role of radiotherapy in the management is unclear and is believed to decrease the rate of local recurrence [5, 6, 15]. Although the role of chemotherapy is debatable, ordinary imaging methods may not be enough to detect the response to chemotherapy. A significant vascular response has been reported in a remarkable percentage of the patients receiving adjuvant chemotherapy [16]. As the LSC is usually metastatic at the time of discovery, palliative resection by wide local excision of the primary tumour may be performed. This procedure should be performed on patients who are not candidates for complete radical resection to improve symptoms, prevent complications, and improve survival [17, 18].

In this article, we present a challenging case of painless scrotal swelling with suspicious radiological criteria that was proven later to be a liposarcoma. Being a rare tumour [1], we aimed to offer this step-wise approach in the diagnosis and post-treatment follow-up of a case of LSC, which might prove useful in future studies concerned with this entity of neoplasms.

2 Case presentation

We present a case of a 53-year-old male who presented to the urology and nephrology center with a large painless right hemiscrotal swelling that showed a progressively enlarging size for 1 year. Scrotal US revealed a large heterogenous soft tissue mass with both hyperechoic and hypoechoic components (Fig. 2). By colour Doppler (CD), the mass showed no significant internal vascularity (Fig. 2). Based on the sonographic findings, the provisional diagnosis was suspicious right hemiscrotal mass. The patient underwent CECT which revealed a large right hemiscrotal lesion extending into the SC showing foci of calcifications with mild post-contrast enhancement (Fig. 3). Another two satellite lesions were synchronously identified in the lower abdomen (Fig. 3). By MRI, the signal intensity of the lesion was low in T1 and high in T2 with no signal drop in fat-suppression sequence (Fig. 4). This signal was atypical for a fat-containing lesion. Based on the radiological findings, the two lower abdominal masses were inoperable as they were adherent to the bowel loops (Fig. 3) and found to be exerting traction on the adjacent mesentery and narrowing of the distal part of the descending and sigmoid colon. There was a subsequent dilatation of the transverse and the proximal descending colon (Fig. 5). The decision was to do a palliative right high inguinal orchiectomy to decrease the tumour burden and refer the patient for postoperative adjuvant chemotherapy.

2.1 Operative details

An inguinal incision was done with an extension to the neck of the scrotum. The scrotum was opened in layers and the cord was identified. The mass was identified and dissected. The cord was ligated. Right orchiectomy was successfully performed and the scrotum was closed in layers.

2.2 Histopathology findings

On gross examination, the operative specimen was the right SC, the testis, and the mass collectively weighing about 1020 g. The SC was 7 cm in length with a mass attached to it measuring about 11×5×5 cm and 2 cm away from the safety margin. The testis was normal in size with the mass attached to it which measured about 13×9×11 cm and 7 cm away from the safety margin. The microscopic examination of the slides prepared from the SC revealed atypical spindle-shaped cells and giant lipoblasts with hyperchromatic nuclei embedded in a dense fibrous stroma (Fig. 6). Slides prepared from the paratesticular mass were also examined revealing fascicles of pleomorphic spindle cells with a focus of peculiar whirling. Immunohistochemical stains for vimentin and S-100 were positive in the SC and negative in the undifferentiated areas in the paratesticular mass. Both were negative for desmin, pan-cytokeratin (Fig. 6) and CD34. The final diagnosis was SC and paratesticular undifferentiated liposarcoma with a free safety margin.

2.3 Outcome

As the adjuvant chemotherapy protocol was highly dependent on the histological type of the residual lesion, the patient underwent an US-guided biopsy from the residual lower abdominal masses. Examination of the slides prepared from the obtained biopsy cores revealed similar pathology. The patient was referred to the medical oncology department for adjuvant chemotherapy and received 6 cycles of doxorubicin-based protocol (doxorubicin and ifosfamide). Sequential follow-up by post-contrast CT after 2 and 6 months revealed a clear right hemiscrotum and a stationary course regarding both the lower abdominal masses and the dilated large bowel loops (Fig. 7).

2.4 Imaging methods

The Doppler ultrasonography examination was performed using both a linear array transducer (7.5 MHZ) and a curvilinear probe (3.5 MHZ) for examination of the scrotum and the SC with a GE-LOGIQ E9 US scanner by a specialist radiologist who had 5 years of experience in uroradiology. The CECT examination of the abdomen and pelvis was performed using a multidetector CT scan

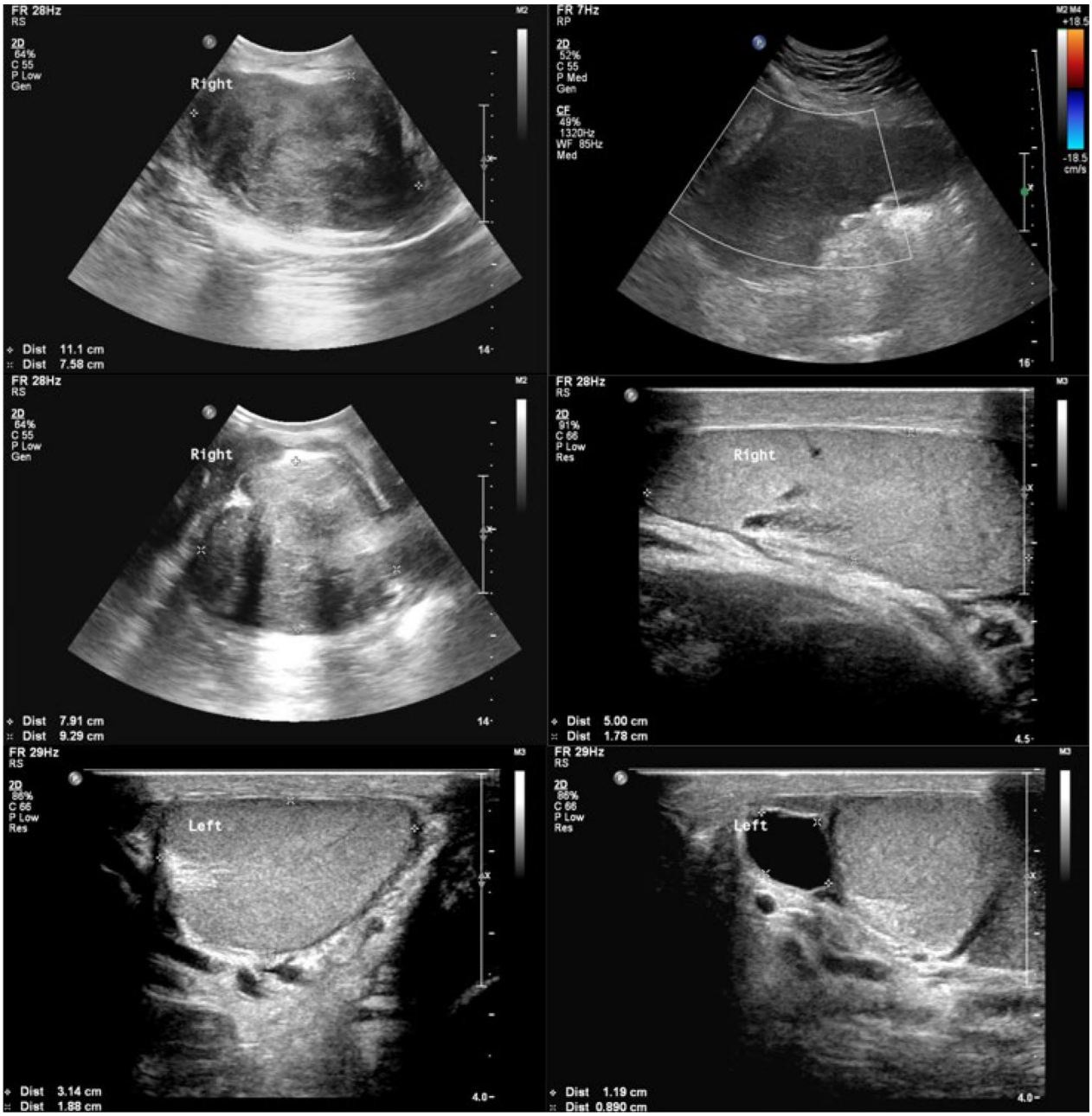

Fig. 2 Testicular and inguinal ultrasound using both linear (7.5 MHZ) and curvilinear (3.5 MHZ) probes: heterogeneous right paratesticular and inguinal masses with mixed hyperechoic and hypoechoic echopattern. No significant internal vascularity by CDUS. Normal left testicle with a small epididymal cyst

(GE-healthcare). The MRI examination of the pelvis and scrotum was performed by a 1.5 Tesla scanner (Philips Ingenia). CT and MRI findings were interpreted by a consultant with 7 years of experience in uroradiology. US-guided biopsy from the lower abdominal masses was performed by a specialist in interventional radiology with 5 years of experience using the US machine mentioned previously and an 18G tru-cut biopsy needle.

3 Discussion

Although liposarcoma is the most prevalent kind of sarcoma in adults [1, 8], LSC is rare [4]. Sambel M. and his colleagues mentioned about 200 LSC cases to have been reported in English written literature [19]. Morozumi et al. mentioned that only 326 cases of LSC have been reported with only 15% of them being of the undifferentiated entity [1].

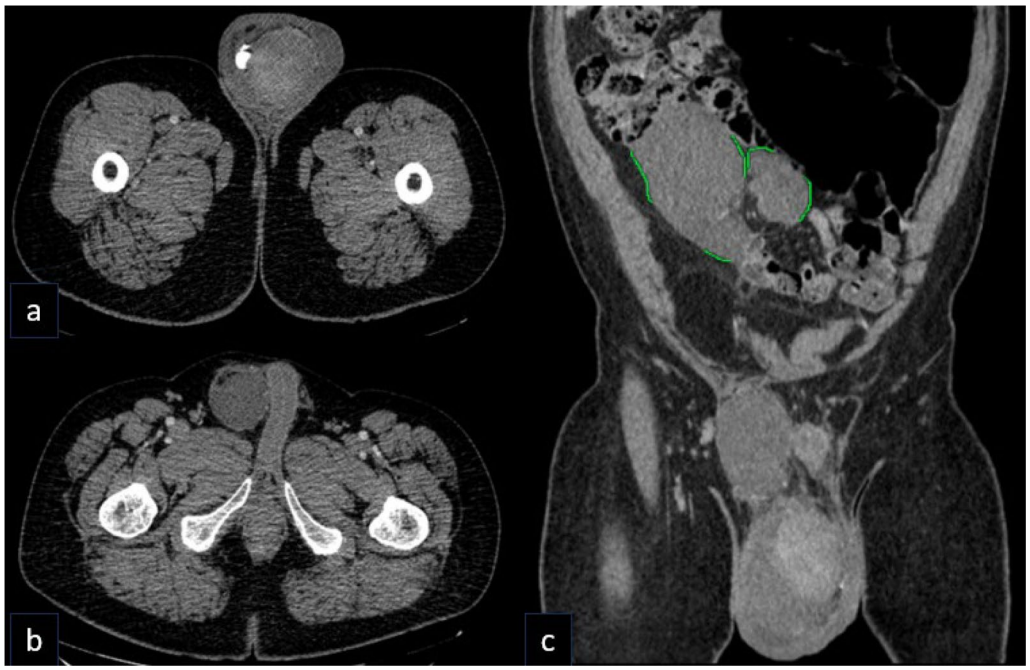

Fig. 3 Post-contrast CT: **a** and **b** axial images: enhancing right paratesticular and spermatic cord masses, respectively, **c** coronal image: enhancing right paratesticular and right inguinal masses. Note the two satellite lesions (green outline) inseparable from the adjacent bowel

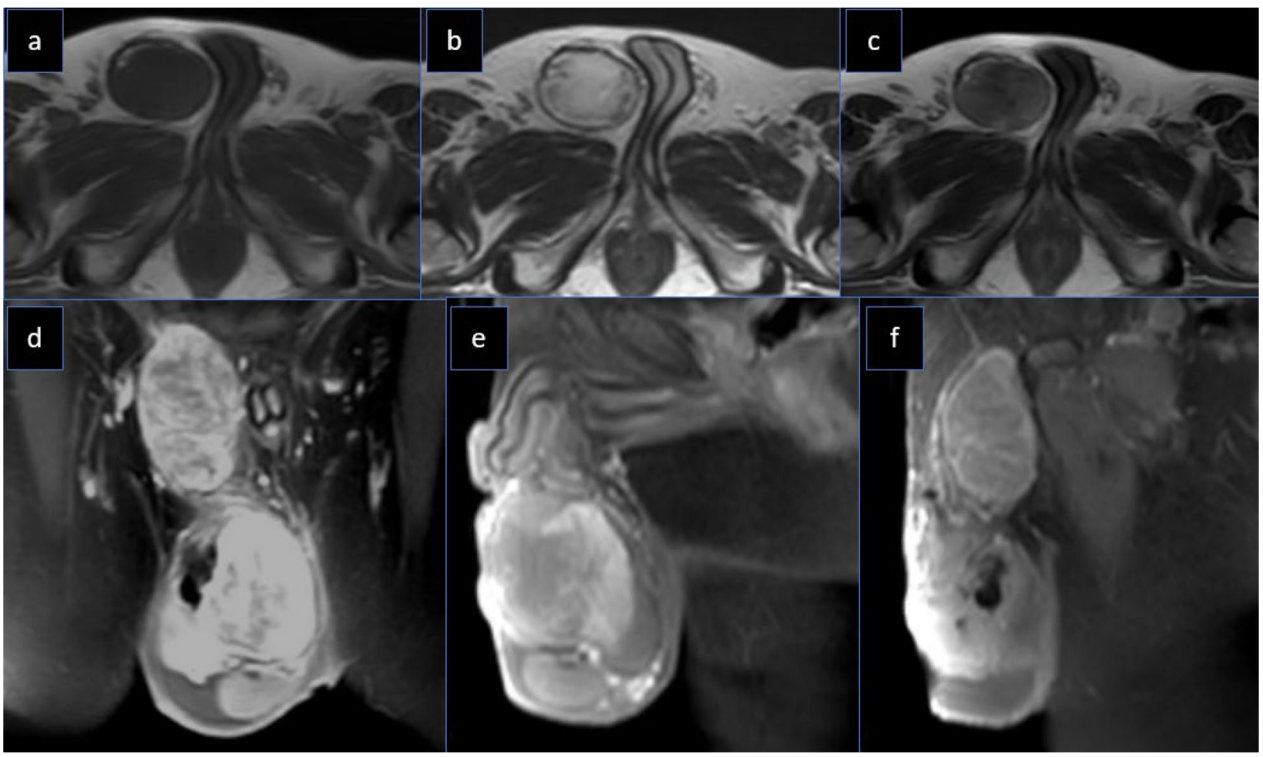

Fig. 4 Pre and Post-contrast MRI: **a**, **b** and **c** axial images: a right spermatic cord lesion displaying low signal in T1 (**a**) and high SI in T2 (**b**) with heterogenous enhancement in T1 with contrast (**c**), **d** coronal FAT-SAT image: enhancing right inguinal and right paratesticular masses showing no signal drop with signal void calcification, **e** and **f** sequential sagittal FAT-SAT images: a paratesticular mass inseparable from the right testicle and an enhancing spermatic cord lesion, respectively

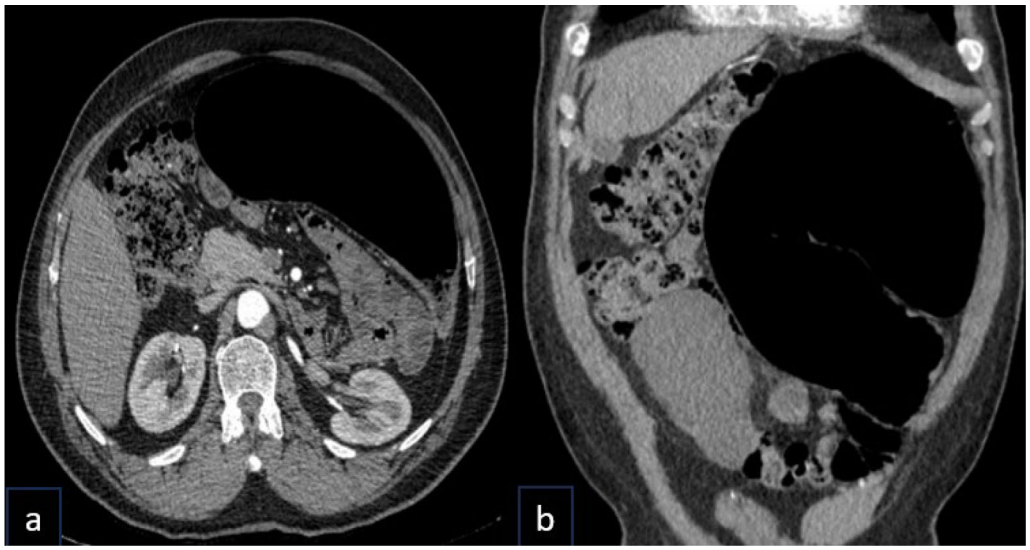

Fig. 5 Post-contrast CT: **a** axial and **b** coronal images: dilated transverse colon and proximal descending colon

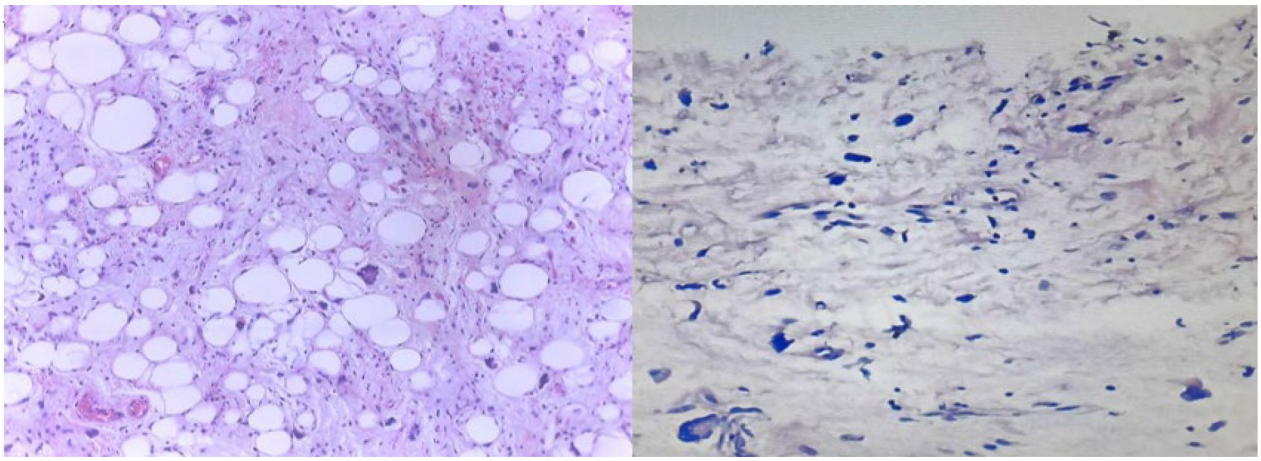

Fig. 6 (Left): hematoxylin and eosin stain [low power] showing infiltration by atypical spindle-shaped cell proliferation, giant cells with hyperchromatic pleomorphic nuclei, and occasional lipoblasts admixed with mature lipocytes on fibromyxoid background. (Right): negative immunohistochemical (IHC) stain for pan-cytokeratin [medium power]

Most of the literature suggests the age of presentation of LSC to be between the 5th and 6th decades of life [20]. Our patient was 53 years old. This agrees with the age range mentioned by Ogbue OD [6] and the case report of Shaban Y, et al., in which the age of the patient in the latter was 59 years [1]. The patient's age in our case report, to some extent, matches that mentioned by Mokrani A., et al. (64 years) [8]. However, Pergel A, et al. mentioned a different age for the patient presented in his case (81 years) [4].

The chief complaint of LSC patients is usually a palpable scrotal or inguinal mass [20]. The presenting symptom in our case was a progressively enlarging inguinal swelling matching the complaint presented by Hamed WMA, et al., Ogbue OD and Shaban Y, et al., who all mentioned similar presentation [1, 6, 7]. Our patient was symptomatic for one year agreeing with Pergel A, et al. who mentioned similar duration with rapid growth in the last 3 months before the time of discovery [4]. On the other hand, Jeremić D, et al. mentioned a shorter period of symptoms (6 months) [12].

Our US description of the lesion matched that of Shimamori N, et al. who described the LSC as a heterogeneous paratesticular mass with mixed echogenicity [21].

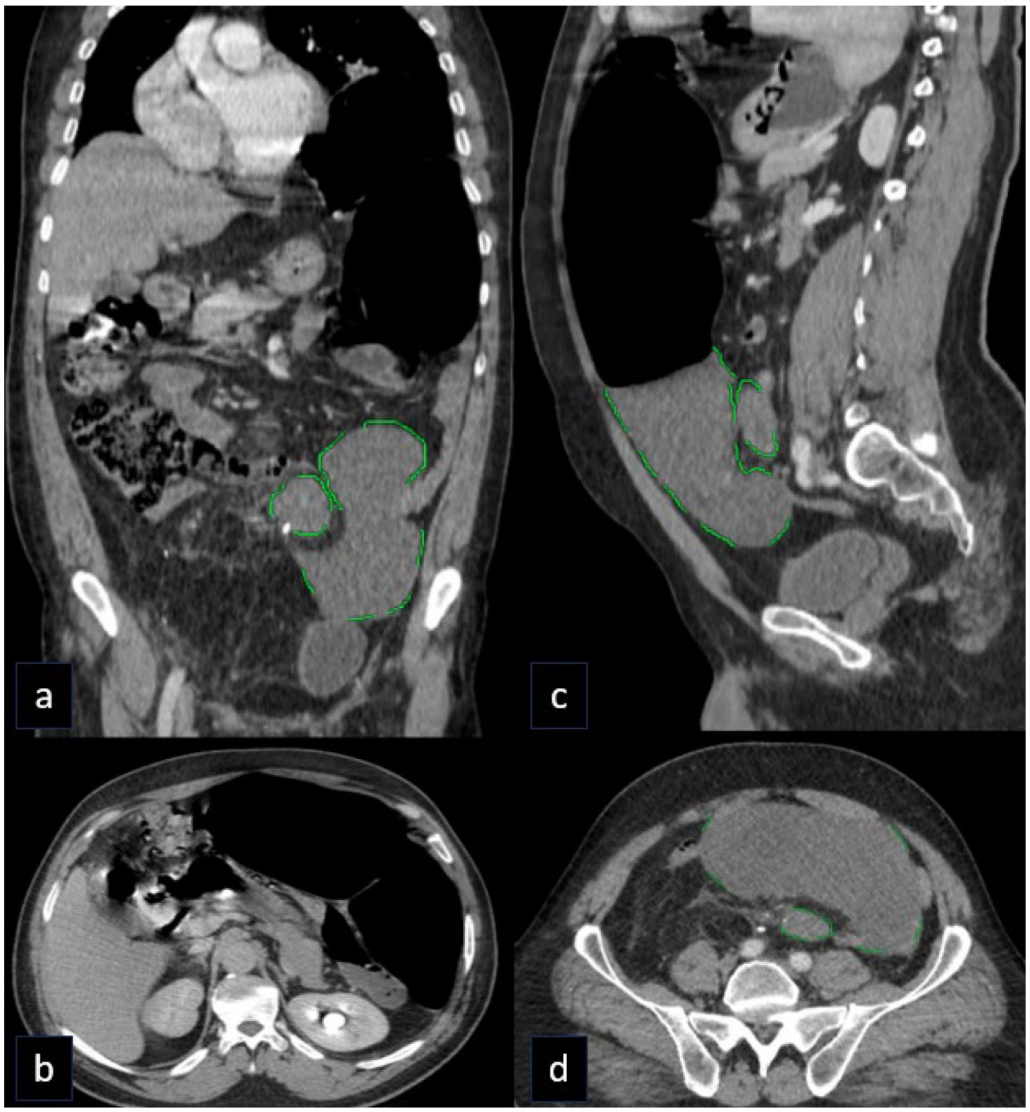

Fig. 7 Post-contrast CT: **a** coronal, **b** and **d** axial, **c** sagittal images. 2-month follow-up [and **b**] and 6-month follow-up [**c** and **d**]: stationary course regarding the satellite lesions (green outline) and the dilated large bowel

Although this description was not specific to LSC, the heterogeneity and indistinct borders were strongly suggestive of a malignant nature regardless of its type by histopathology [21]. Although Jeremić D, et al. described the LSC as a hypervascular mass [12], the lesion in our case showed no significant vascularity by CD. The undifferentiated nature of our lesion might explain this. We concluded that heterogenous paratesticular masses should be considered suspicious regardless of the vascularity of the lesion.

The CECT findings in our case were a large enhancing right hemiscrotal mass with foci of calcifications (Fig. 1). This agrees with the description and enhancement pattern offered by Lu J, et al., who mentioned that the majority of the masses showed islands of flake-like enhancement, reflecting the hypervascular areas within the lesion [22]. In our case, the satellite lesions were discovered by CT synchronously with the original lesion (Fig. 2). This also agrees with Lu J, et al. who mentioned that the presence of satellite lesions was common among the undifferentiated liposarcoma [22]. We concluded that the field of view in the CT examination of a suspicious inguinoscrotal swelling should include the abdomen to reveal remote lesions.

Our lesion showed a low signal in T1 images and a high signal in T2 images by MRI with no significant signal drop in the fat-suppression sequence reflecting the undifferentiated nature (Fig. 4). These findings agree with the

case report of An J. and Kim KW [23]. Henceforth, even in the absence of fat suppression, LSC should be included in the differential diagnosis of the SC lesions. Since the MRI findings in our case were atypical for fat-containing lesions, we concluded that the main role of pre-operative imaging is the accurate delineation of the tumour margins and extension beyond the scrotum rather than the characterization of the nature of the lesion which An J. and Kim KW also concluded [23].

Based on the WHO classification, LSC is categorized into five types well-differentiated, undifferentiated, myxoid, round cell, and pleomorphic [5]. It has been suggested that LSC may arise de novo from SC tissues or on top of malignant transformation of a preexisting lipoma [8]. Although most cases reported in the literature were well-differentiated [12], the histologic subtype of the lesion in our case was found to be undifferentiated. This agrees with the case report offered by Shaban Y, et al. and Mokrani A, et al. who mentioned the presence of a dedifferentiated component [1, 8].

Although surgeons' experience with LSC has been described in many instances, there are no clear management guidelines [1, 8]. Our patient underwent palliative resection with a high inguinal orchiectomy despite the presence of two inoperable satellite lesions at the time of discovery which showed a stationary course on follow-up (Fig. 7). The patients in the cases presented by Shaban Y, et al., Chalouhy C, et al. and Sahnoun W, et al. underwent radical orchiectomy with en bloc resection of the mass [1, 5, 9]. In the case report of Hamed WMA, et al., the lesion was excised with preservation of the spermatic cord and the testicle [7]. Shaban Y, et al., and Chalouhy C, et al. mentioned no recurrence after surgery with no satellite lesions [1, 9]. Hamed WMA, et al. didn't mention the follow-up results of their case [7]. Sahnoun W, et al. mentioned local recurrence after about four years in a patient who had a dedifferentiated component by histopathology [5]. The patient presented by Mokrani A, et al., also underwent orchiectomy, however, the patient was lost to follow-up and subsequently experienced recurrence after two years [8].

The role of adjuvant chemotherapy in LSC remains controversial. The use of adjuvant doxorubicin-based chemotherapy for resected soft tissue sarcomas was evaluated in a meta-analysis and showed an increase in the overall survival [8]. The patient in our case received a doxorubicin-based protocol of chemotherapy. This agrees with Livingston JA, et al. who mentioned that the majority of the patients with liposarcoma received the doxorubicin–ifosfamide protocol [16]. The patient in the case report mentioned by Mokrani A, et al. was also referred for adjuvant chemotherapy [8]. However, in our case, the patient was referred for adjuvant chemotherapy following the primary operation. In the later-mentioned study, the patient was referred for adjuvant chemotherapy following the 2nd operation after recurrence [8].

Finally, LSC is usually mentioned as sporadic cases or as part of larger studies of liposarcoma with no dedicated studies concerned with this specific and rare entity of neoplasms [23]. The diagnosis of LSC is challenging since the presentation is shared by a wide variety of lesions [7, 9, 23]. There is no specific treatment plan and most of the experience is based on trials mentioned in the case reports [1, 8]. Henceforth, we could not compare the adequateness of the patient's response to therapy on follow-up, with those of the other case reports as the course of the disease differed from ours and the follow-up interval was different. Moreover, the lesion in our case showed atypical radiological criteria for a fat-containing lesion owing to its undifferentiated nature. All these factors contributed to the difficulty in our approach to the case.

4 Conclusions

Heterogenous paratesticular masses should be designated suspicious till proven otherwise. Undifferentiated histological types of LSC may show atypical imaging features. The imaging of SC masses should include the abdomen to detect skip lesions. The main role of pre-operative radiology is the accurate delineation of tumour margins. The management of LSC remains controversial with no accepted evidence-based protocol for management.

5 Limitations

The images of the gross specimen were not available, as this article was written retrospectively after reviewing the postoperative radiology. The excised paratesticular and spermatic cord masses were not photographed.

Abbreviations

LSC	Liposarcoma of the spermatic cord
SC	Spermatic cord
CECT	Contrast-enhanced computerized tomography
MRI	Magnetic resonance imaging
US	Ultrasound
CD	Colour Doppler
MHZ	Megahertz
G	Gauge

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Author contributions

All authors have read and approved the manuscript for publishing. MS: analysed the patient's data, wrote the main body of the manuscript, and revised different sections of the document before publishing.IE: provided the patient's data and revised the article before submission. AA: shared in writing the discussion section.AM: shared in the preparation of the figures.SE: revised the case report, provided literature sources, and contributed to the evaluation of radiological findings.

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Availability of data and materials
The data used and/or analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
An informed consent was obtained from the patient.

Consent for publication
An informed consent was obtained from the patient to use the necessary data for publication.

Competing interests
The authors declare that they have no competing interests.

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