



## Oncology

## A case report of giant paratesticular myxoid liposarcoma



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## ABSTRACT

Liposarcomas are an uncommon occurrence in the paratesticular region that makes about 20 % of all sarcomas. The clinical appearance is an inguinal lump, which can resemble a hydrocele or hernia. There would be no conventional treatment accessible because it is such a rare disease. We report the case of a 68-year-old man with paratesticular myxoid liposarcoma. Ultrasound and CT-scan came back in favor of a paratesticular tumor. A high inguinal orchidectomy has been done and the diagnostic of myxoid liposarcoma was first evoked by histology and confirmed by molecular biology. At 12 months follow up the patient remains tumor free.

## 1. Introduction

Sarcomas are a diverse collection of mesenchymal cells-derived malignant neoplasms. Less than 5 % of all soft-tissue sarcomas and less than 2 % of malignant urologic tumors are genitourinary (GU) sarcomas. They can originate from the spermatic cord, inguinal canal, testicular tunic, or epididymis. Initially, they might have been misdiagnosed as an inguinal, scrotal, or hydrocele.<sup>1,2</sup> 30–90 % of GU sarcomas are believed to originate from the spermatic cord, which is the most involved urologic location. Fewer than 350 cases have been reported in the literature.

Although opinions on the best course of action for treating these tumors are still divided, most experts agree that complete resection, including high ligation of the spermatic cord, is the best course of action for all adult patients with peripheral sarcomas.

We present a case report of a giant paratesticular liposarcoma of the right spermatic cord and a comprehensive literature review in order to provide useful information on this rare entity.

## 2. Case report

A 68-year-old man presented for a bulky painless mass of the right inguinal and scrotal region. The patient presented with poor general conditions and a slowly developing tumor that had been present for 18 months. Physical examination showed a giant, firm inguinoscrotal mass causing the left testis and penis to shift laterally (Fig. 1). Serum levels of tumor markers; Beta-Human Chorionic Gonadotropin (HCG), alpha-

fetoprotein ( $\alpha$ -FP), and lactate dehydrogenase (LDH); were normal.

Scrotal Ultrasonography revealed a mixed echogenicity paratesticular mass in contact with the spermatic cord measuring  $17 \times 12,5 \times 8$  cm and vascularized by color Doppler, the right testis and epididymis were both normal, and there was no hydrocele. Thoraco-abdominopelvic CT scan showed a right mixed density paratesticular mass (Fig. 2). No mediastinal, retroperitoneal nor iliac lymphadenopathy was found, and there was no secondary pulmonary or hepatic localization.

The patient underwent a right high inguinal orchidectomy including the mass and all right inguinal canal contents with a high cord ligation (Fig. 3). No postoperative complications were reported.

Macroscopically the tumor appeared as a solid, mucoid mass, without any infiltration into epididymis or testis (Fig. 4). Histology showed a partially necrotic tumor proliferation of spindle-shaped cells with markedly atypical nuclei and a myxoid stroma (Fig. 5). A myxoid liposarcoma has been confirmed by molecular biology.

A 12-month follow-up with thoraco-abdomino-pelvic CT-scan showed no evidence of recurrence. No adjuvant therapy was needed after a multidisciplinary team discussion.

## 3. Discussion

In the literature, there are fewer than 350 cases of paratesticular liposarcomas documented.<sup>3</sup> Roughly 20 % of paratesticular tumors and 5 % of paratesticular sarcomas are liposarcomas. Myxoid liposarcoma is even rarer, representing only 3.3 % of liposarcomas.<sup>1</sup> They are most commonly observed in their 50s and 60s.<sup>3</sup>

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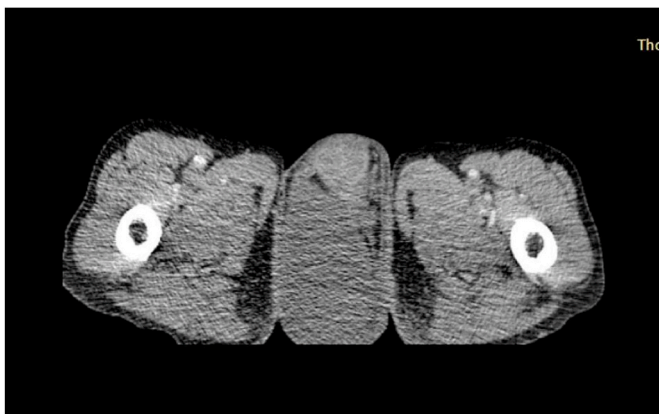
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**Abbreviations:**

GU	Genitourinary
LS	Liposarcomas
US	Ultrasound
MDT	Multidisciplinary discussion team
OS	Overall survival
DFS	Disease-free survival



**Fig. 1.** Huge scrotal mass causing the left testis and penis to shift laterally.



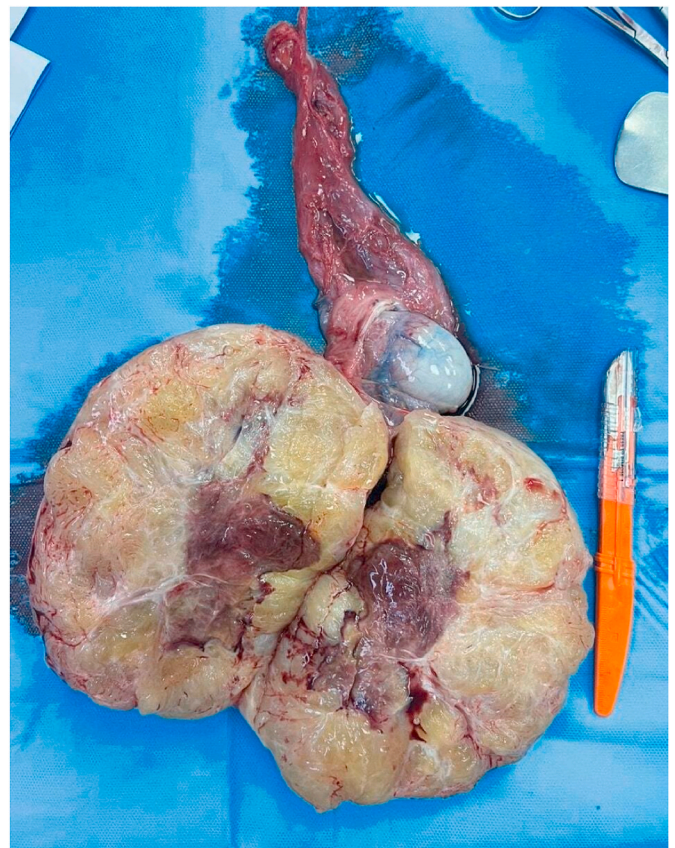
**Fig. 2.** Paratesticular mass with mixed density, is separate from the right testes, with some fat contents within.

There are three primary subtypes of liposarcomas: pleomorphic liposarcoma, myxoid liposarcoma, which now includes “round cell” (high-grade myxoid liposarcoma), and well-differentiated and dedifferentiated liposarcoma, which are recognized as a histologic and behavioral spectrum of one disease entity.<sup>4</sup>

Liposarcomas are similar to those in other organs and can arise in the spermatic cord, scrotum, and epididymis, albeit they are uncommon in the genitourinary tract. They are the most prevalent type of soft-tissue

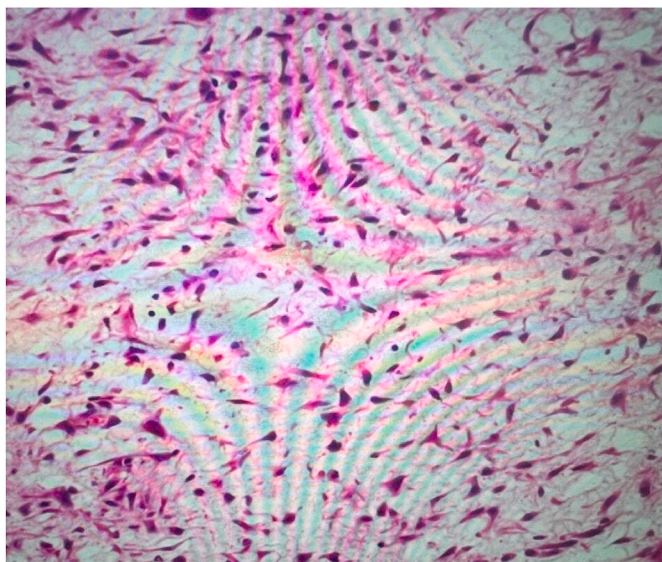


**Fig. 3.** High inguinal orchidectomy including the mass and all right inguinal canal contents.



**Fig. 4.** Macroscopic aspect of the tumor with fatty contours measuring 17 × 12,5 cm. Right testicle appears clearly not invaded.





**Fig. 5.** Microscopic appearance showing a proliferation of spindle-shaped cells with markedly atypical nuclei and a myxoid stroma.

sarcoma in adults. Spermatic cord liposarcomas typically start growing just below the external inguinal ring; hence, as the tumors get big, they appear as painless scrotal masses rather than inguinal masses.<sup>5</sup>

Many times, scrotal LS are misdiagnosed. Particularly in the event of small or well-differentiated tumors with homogenous fatty patterns and moderate development rates that can be mistaken for benign features, physical examination and US are unable to distinguish these entities from lipomas.<sup>6,7</sup> When in doubt, pre-operative CT and/or MRI can offer pertinent information and should always be taken into consideration.<sup>8</sup>

Regardless of their clinical presentation, echogenicity pattern, or growth pace, a diagnosis of paratesticular lymph node syndrome must be considered during the diagnostic workup of scrotal masses due to the possibility of error. Consequently, in these situations, radical orchidectomy combined with upfront extensive local excision is the best course of action. The standard of therapy is generally agreed to be broad resection with ipsilateral orchiectomy and spermatic cord excision; patients with evidence of lymphatic illness should only have locoregional or retroperitoneal lymphadenectomy.<sup>9</sup>

While most paratesticular liposarcomas are primary, some may metastasize to other sites, like the thigh or the fatty tissue around the testicle. Its intermediate malignant nature is the cause of its local recurrence; distant organ metastases are uncommon. With spermatic cord liposarcoma, recurrences occur in 1/4 of patients, and metastases through lymphatic or hematogenous spread occur in 1/10 of patients. Consider adjuvant radiation covering the inguinal area and scrotum if an adequate tumor-free margin cannot be reached.<sup>5</sup> Because of the unusually high probability of local recurrence and distant metastasis, paratesticular liposarcomas always require long-term follow-up, regardless of potential risk factors or the type of treatment taken.<sup>10</sup>

Positive surgical margins appear to be the primary risk factor for both distant metastases and early local recurrence. Microscopic residual disease has been seen in 27 % of cases of seemingly complete excision following wide excision.<sup>11</sup> If the margins are positive, it is advised to perform an even wider second resection.<sup>12</sup> Rather, there is ongoing debate on the usefulness of adjuvant medicines such chemotherapy and radiation.<sup>13</sup> Since liposarcomas are the most radiosensitive of all sarcomas, radiotherapy by itself has been used to induce remission in a few cases.<sup>14</sup> While there was no gain in OS, two randomized trials did demonstrate a decrease in DFS and local recurrence.<sup>14</sup> As a result, radiotherapy may be advised in cases of aggressive high-grade tumors, lymphovascular invasion, or positive margins discovered during the definitive histological examination. It may also be helpful for control in

the event of local recurrences. However, there are currently no data regarding the use of adjuvant or neo-adjuvant chemotherapy in the management of LS.

In our case, the patient underwent a high inguinal orchidectomy, and there was no tumor visible on the surgical margins. Twelve-months following radical surgery there was no evidence of tumor recurrence or metastases.

To sum up, preoperative diagnoses of myxoid liposarcoma are frequently mistaken. The optimum course of treatment is a radical orchiectomy with extensive local excision and high ligation if the condition is identified or highly suspected prior to surgery; adjuvant radiation is advised if the margin status is unclear. Given the possibility of distant metastases and local recurrence, long-term follow-up is advised.<sup>15</sup>

#### 4. Conclusion

Rare tumors called paratesticular liposarcomas are frequently misdiagnosed before surgery. The diagnosis is aided by CT, MRI, and ultrasound scans. However, histological, immunohistochemical, and molecular genetic investigations serve as the basis for the confirmative diagnosis. When there is a clinical suspicion, a high inguinal orchidectomy with local excision should be performed right away. Improving illness outcomes requires frequent follow-ups along with MDT meetings.

#### CRedit authorship contribution statement

**Jaafar Fouimtizi:** Writing – review & editing, Writing – original draft, Data curation. **Youssef Maachi:** Writing – original draft. **Anass Rhayour:** Writing – original draft. **Amine EL Boustani:** Writing – original draft. **Amine Slaoui:** Writing – review & editing. **Tariq Karmouni:** Writing – original draft. **Khalid EL Khader:** Writing – review & editing. **Abdellatif Koutani:** Writing – review & editing. **Ahmed Iben Attya Andaloussi:** Writing – review & editing.

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