ACS Case Reviews in Surgery
Vol. 4, No. 7

Extratesticular Mesenchymal Liposarcoma Found During Inguinal Hernia Repair Surgery in a 65-Year-Old Male

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Background	A 65-year-old male presented with physical exam findings suggestive of an inguinal hernia and underwent open hernia repair, resulting in the discovery of a mass, with final pathology revealing extra-testicular mesenchymal liposarcoma (ETML).
Summary	Our patient presented with a left scrotal bulge that had been progressively worsening over several months. Physical examination revealed a significant left inguinal canal bulge extending into the left scrotal sac. Ultrasound of the area showed an adipose-containing inguinal hernia with a smaller left testicle compared to a previous ultrasound obtained five years earlier. Open surgical repair of the hernia was performed, revealing a large mass that was subsequently excised. Postoperative pathology of the mass was consistent with ETML. Due to their rarity (less than 200 reported cases), ETMLs are frequently misdiagnosed preoperatively. The associated symptomatology, like increased prominence with Valsalva maneuvers, can further mimic an inguinal hernia, contributing to diagnostic challenges.
Conclusion	This case emphasizes the importance of maintaining a broad differential diagnosis when evaluating scrotal masses. ETML, though rare, should be considered alongside more typical diagnoses like inguinal hernia.
Key Words	liposarcoma; testicular; hernia

DISCLOSURE STATEMENT:
The authors have no relevant financial relationships to disclose.

FUNDING/SUPPORT:
The authors have no relevant financial relationships or in-kind support to disclose.

RECEIVED: May 12, 2023
REVISION RECEIVED: December 9, 2023
ACCEPTED FOR PUBLICATION: January 24, 2024

To Cite: Clemons TH, Lopez-Flores RD, Stumpf P. Extratesticular Mesenchymal Liposarcoma Found During Inguinal Hernia Repair Surgery in a 65-Year-Old Male. *ACS Case Reviews in Surgery.* 2024;4(7):21-25.

Case Description

Extra-testicular mesenchymal liposarcoma (ETML) is a rare sarcoma with less than 200 reported cases internationally.[1] It typically presents in adults over 50 years old[2] with clinical features including a bulge in the affected area that can be painless or may be accompanied by swelling, erythema, or warmth. Only 10% of cases originate in the testicular sac, and even fewer reach sizes close to 10 cm.[3] Due to its rarity, data on the incidence, natural history, diagnosis, and treatment of ETML remains limited.

A 65-year-old Caucasian male with a past medical history of gastroesophageal reflux disease (GERD), hyperlipidemia, and a previous left spermatocelectomy, presented to the Veteran's Affairs (VA) clinic in 2022 for surgical consultation regarding an inguinal hernia. The patient reported no history of smoking and occasional alcohol consumption.

In 2016, he presented to primary care with a complaint of straining following lifting and a noticeable bulge in his left inguinal area. Following up with urology in 2017 due to increasing discomfort from the bulge, he was diagnosed with a spermatocele. In 2017, the patient had a spermatocelectomy, at which time a large spermatocele was removed from his left testicle. Operative notes during this procedure indicate that the testicle was completely visualized and had no indication of malignancy.

In August 2022, the patient presented to a urologist with a small left scrotal bulge that had rapidly increased in size after heavy lifting. He thought it may be a recurrent spermatocele. Physical exam was notable for scrotal swelling consistent with an inguinal hernia.

A scrotal ultrasound was performed to differentiate between a recurrent spermatocele and an inguinal hernia. The radiologist interpreted the ultrasound as a large, adipose-containing inguinal hernia (Figure 1). No scrotal lipoma or suspicious adipose mass was identified on ultrasound, so no further imaging was suggested. Additionally, the ultrasound noted atrophy of the left testicle compared to a previous ultrasound performed in 2016 (Figure 2). Based on these findings, the patient to our office for surgical repair of a presumed inguinal hernia.

Figure 1. Scrotal Ultrasound of Testicular Mass (left inguinal view). Published with Permission.

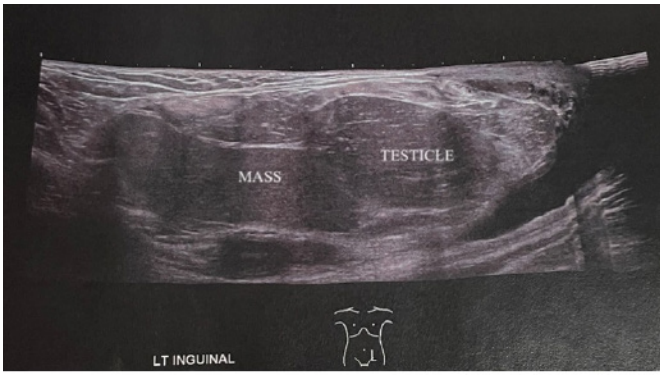
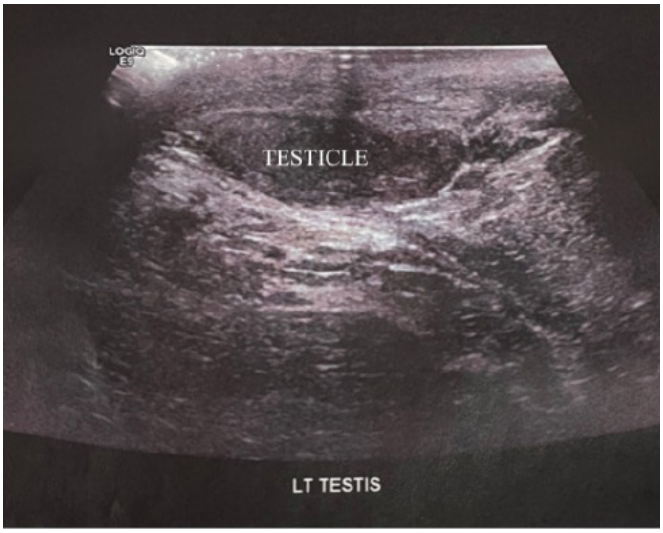


Figure 2. Scrotal Ultrasound of Testicular Mass (left testicular view). Published with Permission.



The patient reported only pressure and discomfort in the scrotum. The only subjective difference from a typical inguinal hernia was the absence of enlargement with straining or heavy lifting. Physical examination revealed a full left scrotum with a non-reducible hernia. No other abnormal findings suggestive of a diagnosis beyond a large inguinal hernia containing adipose tissue were detected.

No preoperative interventions were necessary. The patient underwent open inguinal hernia repair in September 2022. A standard surgical approach was employed, involving a transverse incision across the inguinal canal. The external

and internal oblique muscles were exposed, shelf areas were developed bluntly, and the spermatic cord was digitally manipulated to bring it into the operative field. Despite a weak abdominal wall being present, neither a typical direct inguinal hernia sac nor an indirect inguinal hernia sac was visualized. However, an additional mass was present in the scrotum. Gentle retrograde pressure delivered a large lipomatous mass into the inguinal canal, encasing the testicle. The mass was meticulously dissected free from the testicle. Intraoperative consultation with urology was obtained, and due to diagnostic uncertainty, the urologist advised against testicular removal at that time. Depending on pathology results, a potential salvage orchiectomy was discussed.

Once the mass was removed, the abdominal wall was repaired with a 10 × 15 cm partially absorbable mesh. The patient tolerated the procedure well and was discharged home. At a two-month follow-up, the patient reported no pain except with direct palpation of the testicle. Exam showed mild induration and tenderness of the spermatic cord and testicle.

Initial pathology results described a homogenous, tan lesion with a consistency resembling fish flesh, measuring 9 × 9 × 6 cm (Figure 3). Subsequent surgical pathology identified the mass as a lipomatous tumor containing lipoblasts (Figure 4). The specimen was then sent to an external pathology lab for further characterization. This analysis revealed grossly positive margins for the mass, which was categorized as an atypical lipomatous tumor/well-differentiated liposarcoma (ALT/WDLS) based on molecular markers. These markers included:

- MDM2 gene amplification: Helps differentiate liposarcoma from benign lipoma.
- MDM2/Cap12 ratio greater than 4.6: A ratio exceeding 3.0 supports the diagnosis of ALT/WDLS.

Following the final pathology read of the mass obtained in October 2022, the patient was referred to oncology for follow-up CT imaging to assess for potential retroperitoneal invasion and to urology for a possible radical orchiectomy. A general surgery consultation at the VA in November 2022 found no acute complaints. The patient was subsequently referred to another VA facility for presurgical consultation in December 2022 and scheduled for an orchiectomy in January 2023. However, concerns for retroperitoneal involvement and the need to remove the hernia repair mesh prompted the urology team to consult with general surgery before proceeding with the orchiectomy.

Figure 3. Gross Examination of Small Section of ETML. Published with Permission.

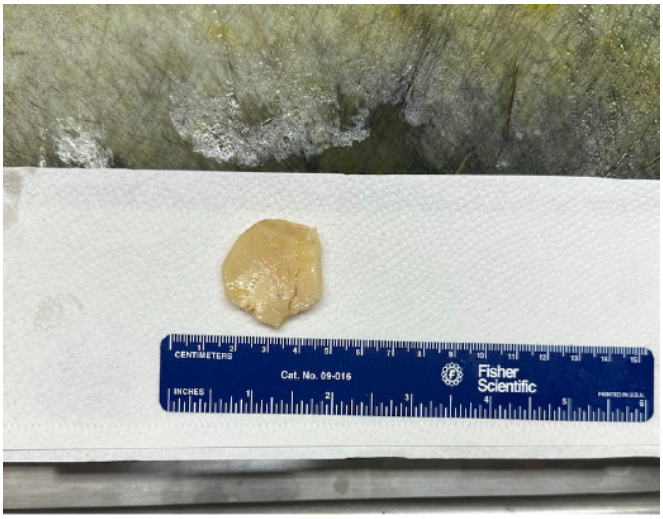
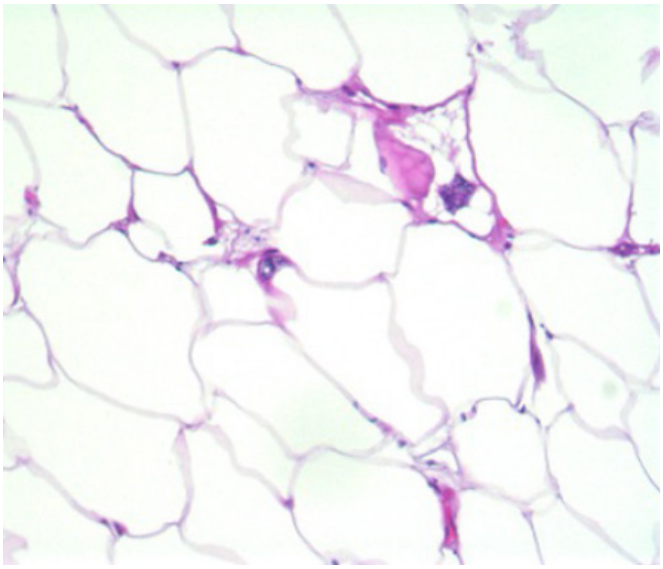


Figure 4. Histopathology of ETML Showing Lipoblasts. Published with Permission.



After discussion, the general surgery team recommended a course of surveillance CT scans every six months with the option of a salvage orchiectomy if recurrence was noted. The patient was informed of this plan and expressed his preference for surveillance over immediate orchiectomy. A surveillance CT scan in January 2023 showed no evidence of recurrence, with the next scan planned for August 2023.

Discussion

This case report details an exceptionally rare presentation of an extra-testicular mesenchymal liposarcoma (ETML)[3] originating from the testicle and reaching nearly 10 cm in size. While ETMLs typically affect adults in their 50s-60s (range 16-87 years),[1,2] their presentation is usually localized with a low metastatic rate (around 4% at diagnosis).[4]

A significant challenge in diagnosing ETML lies in its potential to mimic other conditions, as seen in this case where a preoperative diagnosis of an adipose inguinal hernia was made. This is likely due to our patient's limited imaging workup, which only included an ultrasound. On ultrasound, smaller ETML tumors often mimic lipomas or appear as homogenous fat deposits. While larger tumors may exhibit a more heterogeneous appearance with hypoechoic regions and necrosis, this case demonstrates that even tumors approaching 10 cm may maintain a homogeneous, lipomatous presentation.

Even with magnetic resonance imaging (MRI), which can differentiate space-occupying lesions within the testicle from hernias, misdiagnosis can occur. Prior cases have reported mistaking ETML for spermatocytoma.[5] In this instance, an MRI might have prompted a radical orchiectomy upfront due to the concern for seeding with a testicular tumor biopsy. However, the rarity of this tumor makes routine MRI use for inguinal masses impractical in similar cases with a low index of suspicion for a tumor.

Furthermore, distinguishing ETML from benign lipomas can be difficult based solely on imaging and gross pathology, potentially leading to incorrect preoperative diagnoses. Definitive diagnosis requires special staining techniques, as MDM2 and CDK4 immunostaining are the key markers that differentiate liposarcoma from benign lipomas.[2]

Due to the rarity of ETML, survival and treatment data have not been well explored in the literature. The most frequent treatment appears to be radical orchiectomy with high ligation of the spermatic cord at the inguinal ring. Incomplete excision is associated with an increased recurrence rate. A recent study found that while three-year progression-free survival was 100% for removal with negative margins, this number drops to 29% if there are positive margins.[4] Another study revealed the five-year survival rate with excision was 75%, and the recurrence rate was 50-70% of all cases.[6] In this patient, he had not received informed consent about the possibility of an orchiectomy, so if the mass had been clearly malignant, the radical orchiectomy would have been inappropriate at that time, regardless of the data on recurrence.

The rapid growth of the tumor, as evidenced by its absence during a previous examination five years prior, along with its significant size and symptomatic presentation, strongly suggested a malignant process. While well-differentiated liposarcomas are generally slow-growing, this case demonstrates their potential for rapid expansion.

Given the probable diagnosis of ETML, radical orchiectomy would be the recommended approach. However, in this specific case, the patient had not been informed of this possibility preoperatively. Therefore, radical orchiectomy was not a viable option, despite the potential benefits in terms of reducing the risk of recurrence.

As an alternative, regular surveillance with CT scans every six months is recommended to monitor for any signs of recurrence. If evidence of recurrence emerges, salvage orchiectomy can be considered as a subsequent step.

Conclusion

Extratesticular mesenchymal liposarcoma (ETML) is a rare type of cancer that can be easily misdiagnosed as an inguinal hernia. This case report presents a patient with a large ETML that developed within five years of previous testicular surgery and was initially mistaken for a hernia based on clinical presentation and imaging findings. This case highlights the importance of maintaining a broad differential diagnosis when evaluating testicular masses.

Lessons Learned

Inguinal masses, whether caused by tumors or hernias, can present with similar histories, physical exam findings, and imaging characteristics. A definitive diagnosis often requires surgical exploration and comprehensive pathological examination of the tissue.

Acknowledgment

Drs. Grant Hayashi and John Laughlin were responsible for assistance with image acquisition, and Dr. Bradley Bandera was responsible for editorial support. The University of Nevada, Reno School of Medicine Open Access Fund provided partial funding for open access publication.

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