Submitted: 17 February, 2025 Accepted: 25 August, 2025 Published: 30 November, 2025 DOI:10.22514/jomh.2025.136

CASE REPORT

JOMH Journal of Men's Health

Malignant biphasic mesothelioma of the tunica vaginalis testis: a case report

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Abstract

Background: Malignant mesothelioma of the tunica vaginalis testis (MMTVT) is a rare malignancy often presenting with non-specific symptoms, leading to diagnostic challenges. **Case**: We report the case of a 36-year-old patient initially presenting with hydrocele-like symptoms, highlighting the complexities of MMTVT diagnosis and management. Furthermore, while asbestos exposure is a recognized risk factor, this case lacked a significant history of such exposure, suggesting the need to explore alternative etiological factors. **Conclusions**: Radical orchiectomy remains the standard treatment, yet the role of adjuvant therapies is not yet well-defined due to the rarity of the disease.

Keywords

Malignant mesothelioma of the tunica vaginalis testis (MMTVT); Asbestos exposure; Hydrocele; Magnetic resonance imaging; Computertomography scan; Positron emission tomography CT; β-human chorionic gonadotropin (β-HCG); Alpha feto-protein (AFP); On-bloc hemiscrotectomy

1. Introduction

Malignant mesothelioma is a rare tumor predominantly associated with asbestos exposure, a connection first documented by Wagner *et al.* [1] in 1960. This neoplasm can arise from the mesothelial cells lining the pleural, peritoneal, and pericardial cavities, and the tunica vaginalis testis. Among the various types of mesothelioma, malignant mesothelioma of the tunica vaginalis of the testis is particularly rare, accounting for approximately 0.3 to 5% of all mesothelioma cases, as highlighted by Iczkowski in 2023 [2]. This form of mesothelioma presents unique diagnostic and therapeutic challenges, attributed to its rarity and the non-specific nature of its clinical presentation. The limited number of cases reported in international publications, fewer than 300 since the initial documentation by Barbera and Rubino in 1957, reflects this rarity [3]. The current case report aims to contribute to the existing literature by detailing the diagnosis, treatment, and follow-up of a rare case of malignant biphasic mesothelioma of the tunica vaginalis testis, underscoring the importance of considering this diagnosis in patients with testicular masses and hydrocele, especially those with a history of asbestos exposure.

2. Case presentation

In March of 2023, a 36-year-old male with no significant past medical history presented to an external urology department with a chief complaint of sudden onset of painless swelling of the right hemiscrotum. The patient reported intermittent night sweats for four years, excessive sweating without physical activity, and occasional pain in the right inguinal region. He had no known pre-existing conditions, urological interventions, or family history of cancer.

A scrotal ultrasound was performed as the primary diagnostic step, which confirmed a significant 6 $\times$ 5 cm right hydrocele. The key finding, however, was a distinct 16 $\times$ 13 mm exophytic mass arising from the tunica vaginalis. This mass was described as having a heterogeneous echotexture in the imaging.

Based on these imaging findings, the differential diagnosis included both benign and malignant entities, such as a reactive inflammatory process, an adenomatoid tumor, or a rare tumor of the tunica vaginalis. Given the atypical appearance of the mass, the clinical suspicion for a malignant process was high.

To avoid the risk of tumor seeding, a percutaneous biopsy was not appropriate. Consequently, an upfront surgical approach was chosen. The patient underwent a hydrocelectomy and concurrent excision of the hydrocele wall mass for definitive histopathological diagnosis. This surgical strategy aimed to achieve both symptomatic relief and a complete, one-step diagnostic and therapeutic intervention. The histopathological evaluation later identified malignant biphasic mesothelioma of the tunica vaginalis testis. Subsequent investigations, including abdominal Megnetic Resonance Imaging (MRI) and Computertomography (CT) scan, showed no evidence of metastasis but revealed incidental findings of a secondary spleen and small cysts in the left liver lobe and left kidney. Laboratory results for Thyroid Stimulating Hormone (TSH), β-Human Chorionic Gonadotropin (β-HCG), and Alpha Feto-Protein

(AFP) were within normal limits.

The patient was then referred to our department and underwent on-bloc hemiscrotectomy on the right side, including ablation of the right testis and the spermatic cord (Fig. 1). The perioperative course was uneventful, and the patient was discharged with a plan for follow-up and further imaging. The histopathologic examination confirmed malignant mesothelioma of the Tunica Vaginalis Testis (MMTVT, Fig. 2) with no infiltration of the testicle or epididymis. Spermatogenesis appeared normal. There was no evidence of a tumor at the resection margins of skin or spermatic cord.

The patient's follow-up was conducted in an outpatient setting through a locally based urological practice with a special focus on uro-oncology. Both clinical findings and laboratory parameters, as well as ultrasound and MRI diagnostics, remained normal throughout the course. As a part of further follow-up, the patient will continue to be managed within the practice.

3. Discussion

The rarity, non-specific presentation, and complex etiology of MMTVT pose significant challenges in its management and understanding. The present case significantly advances our understanding of MMTVT. Its analysis alongside existing literature demonstrates its occurrence in a remarkably young patient (36 years old) and highlights the diagnostic challenges despite advanced imaging modalities.

MMTVT often presents as benign conditions such as hydrocele or inguinal hernia, leading to diagnostic delays. This is in line with the findings by Segura-González *et al*. [4] (2015), who noted that a significant number of cases were diagnosed during surgeries intended for presumed benign conditions. The case clearly highlights the diagnostic ambiguity, presenting initially with hydrocele-like symptoms without suspicion of malignancy. Clinicians must maintain a high index of suspicion for MMTVT, especially in patients with unusual or persistent scrotal pathology.

Literature indicates a clear predisposition for MMTVT in males between 55 and 75 years, a finding confirmed by Plas *et al*. [5] (1998) and Butnor *et al*. [6] (2019), who reported a median age of 72 years. However, this 36-year-old patient's occurrence is a significant deviation from the typical age profile, substantially broadening the known age spectrum. It is vital to include MMTVT in differential diagnoses, even for younger patients. Age should not be used as an exclusionary criterion.

Asbestos exposure has been identified as a significant risk factor for the development of mesothelioma, including MMTVT. While Plas *et al*. [5] (1998) observed asbestos exposure in a third of their cases, our case lacked a significant history of asbestos contact, highlighting the possibility of other etiological factors contributing to MMTVT development.

This observation is consistent with the broader understanding that while asbestos remains a significant risk factor, MMTVT can occur independently of known

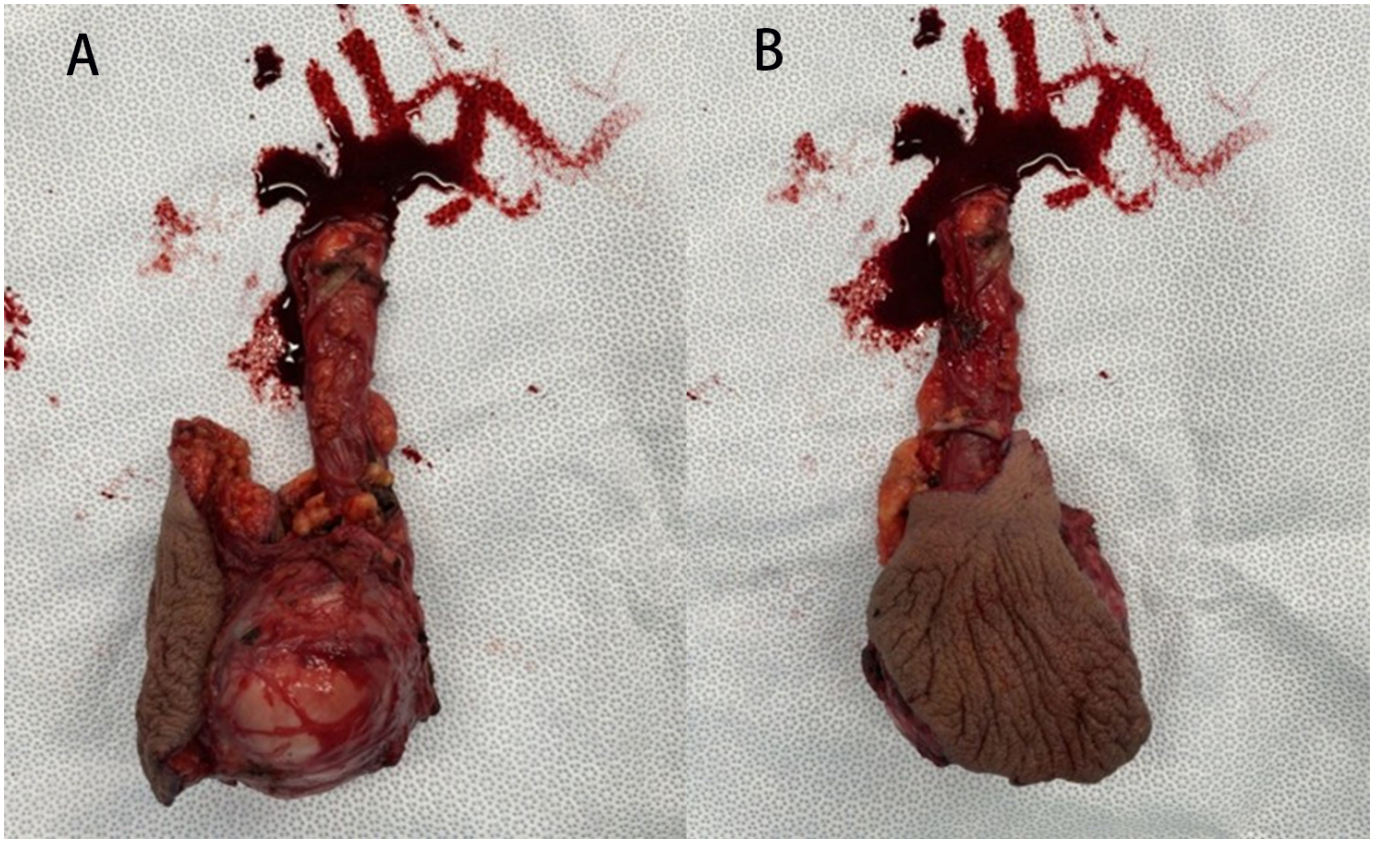

FIGURE 1. Macroscopic anterior (A) and posterior (B) views of the surgical specimen following right-sided hemiscrotectomy, including the testis, spermatic cord, and tunica vaginalis. The exophytic tumor is visible on the hydrocele wall.

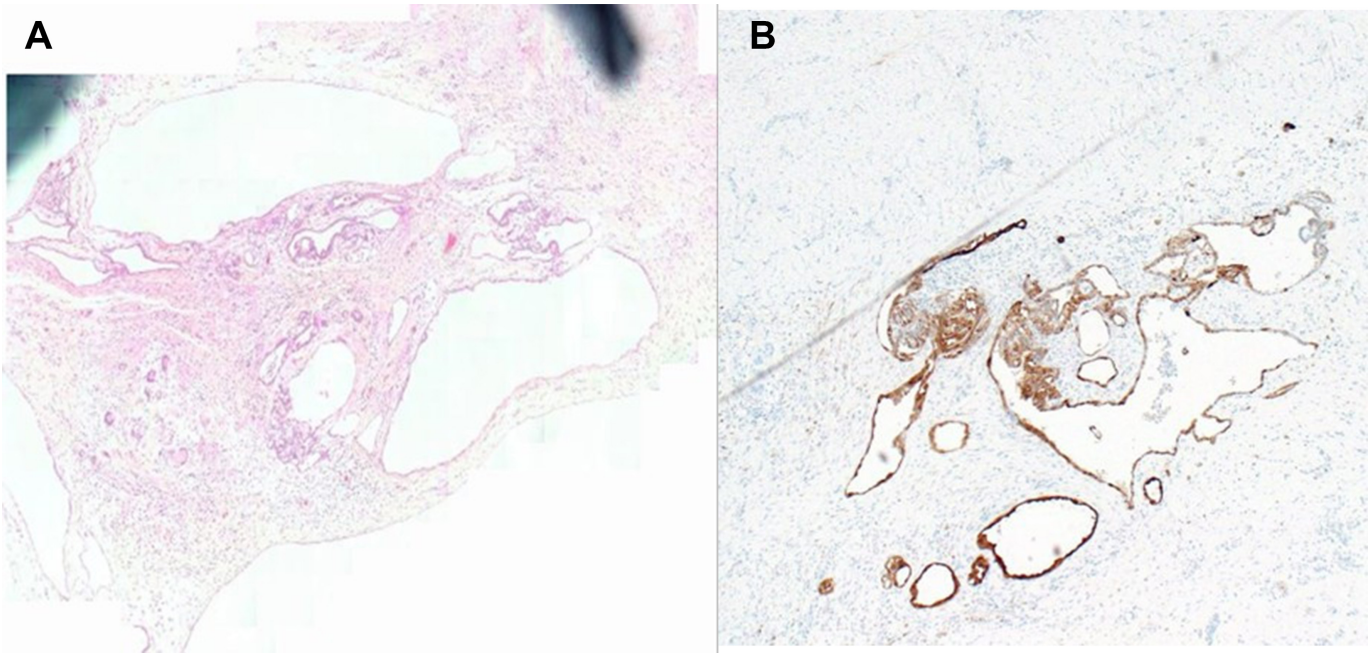

FIGURE 2. Histopathological and immunohistochemical findings of MMTVT. (A) Hematoxylin and eosin (H&E) staining at 10× magnification showing biphasic tumor architecture with both epithelioid and sarcomatoid components. (B) Immunohistochemical staining for Calretinin (10× magnification), demonstrating strong cytoplasmic and nuclear positivity in tumor cells, consistent with mesothelial origin.

asbestos exposure, necessitating further exploration into its pathogenesis.

The absence of a gold standard for radiologic imaging in MMTVT poses significant diagnostic challenges. Although MRI, incorporating Diffusion Weighted Imaging (DWI) sequences, was conducted on the patient, no restricted diffusion indicative of malignancy was detected. This finding, though atypical for some aggressive malignancies, highlights the variability in MMTVT imaging presentation and the limitations of conventional MRI in definitively differentiating benign from malignant scrotal pathologies. The decision not to perform an initial Positron Emission Tomography CT (PETCT) scan was based on the absence of symptoms or imaging suspicion for peritoneal involvement at the time of presentation. However, given the propensity of MMTVT for local invasion and potential peritoneal dissemination, PETCT is widely considered crucial for comprehensive staging and detection of occult metastatic disease, even if performed later in the diagnostic workup or for surveillance.

Radical orchiectomy remains the cornerstone of MMTVT treatment. In this particular instance, given the absence of metastatic disease and complete surgical resection with negative margins (R0 resection), the decision was taken not to initiate adjuvant chemotherapy or targeted therapy. This decision was primarily based on the lack of established adjuvant treatment protocols for MMTVT due to its rarity, as well as the current expert consensus and existing case series that favour surgery alone in non-metastatic cases.

Segura-González *et al*. [4] (2015) advocated for a multimodal approach in patients with locoregional disease, suggesting the potential benefit of integrating surgery, chemotherapy, and radiotherapy. Furthermore, the treatment recommendation for our patient reflected the current practice which favors monotherapy in completely resected early-stage disease.

In our performed histopathological diagnostics, there was cell- and nuclear atypia and invasive growth thus establishing the diagnosis of malignant mesothelioma. This obviates the necessity of any other stainings like Kiel (KI)-67 which is frequently used in oncology to estimate a tumor's proliferation index.

This case, set against the backdrop of current literature, reinforces the need for vigilance and a multidisciplinary approach in diagnosing and managing MMTVT. It underlines the diagnostic challenges posed by its unspecific presentation and the critical need for awareness among clinicians. Future research should aim to elucidate the full spectrum of etiological factors, improve diagnostic methodologies, and refine treatment modalities to enhance outcomes for patients with MMTVT.

This case report has several limitations. As an isolated observation, it does not allow generalization of diagnostic or therapeutic conclusions for malignant biphasic mesothelioma of the tunica vaginalis testis. The rarity of the disease and the lack of standardized treatment protocols further restrict comparability with other cases. In addition, limited follow-up reduces the ability to comment on long-term prognosis or recurrence. Finally, diagnostic challenges inherent to biphasic mesothelioma and incomplete clinical information—such as detailed exposure history or molecular data—may limit the depth of interpretation.

4. Conclusions

In conclusion, the present case of MMTVT in a 36-year-old patient critically expands our understanding of this rare ma-

lignancy. It particularly emphasizes the importance of clinical vigilance even in atypical patient demographics and highlights the challenges of diagnostic imaging, underscoring the need for comprehensive evaluation and management strategies.

AVAILABILITY OF DATA AND MATERIALS

All data generated or analyzed during this study are included in this published article.

AUTHOR CONTRIBUTIONS

LK and MeH—contributed equally to this work. LK and AS—designed the research study. MeH—performed the research. PS, SS, CS and MiH—provided help and advice on patients data. LK and DF—wrote the manuscript. MF and AS—critically reviewed the manuscript, provided guidance on the analysis, and approved the final version for publication. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Our urology clinic at the university medical center Hamburg-Eppendorf does not require ethical approval for reporting individual cases. Consent form was obtained from the participant.

ACKNOWLEDGMENT

The authors would like to thank the patient for providing consent to share the details of this case.

FUNDING

This research received no external funding.

CONFLICT OF INTEREST

The authors declare no conflict of interest. Armin Soave is serving as one of the Editorial Board members of this journal. We declare that Armin Soave had no involvement in the peer review of this article and has no access to information regarding its peer review. Full responsibility for the editorial process for this article was delegated to BB.

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How to cite this article: Lennart Kühnke, Melisa Hoshaber, Stefan Steurer, Philipp Schriefer, Margit Fisch, Dejan Filipas, *et al*. Malignant biphasic mesothelioma of the tunica vaginalis testis: a case report. Journal of Men's Health. 2025; 21(11): 64-67. doi: 10.22514/jomh.2025.136.