Case Report

Clinical Features and Outcomes of Tunica Vaginalis Mesothelioma: A Case Series From the National Institutes of Health

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Clinical Practice Points

- Mesothelioma of the tunica vaginalis is a rare malignancy presenting often as a painless testicular mass.
- Previous reported cases have described poor prognosis of tunica vaginalis mesothelioma despite aggressive surgery and systemic therapy.
- Aggressive surgical management should be first-line treatment because more conservative options might increase the risk of recurrence.
- There are currently limited data to support the benefit of adjuvant therapies such as radiation or chemotherapy.
- Post-treatment surveillance is also imperative and should include imaging routinely within the first 2 years.
- Negative asbestos exposure screening during history should not eliminate clinical suspicion.

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Introduction

Malignant mesothelioma of the tunica vaginalis is an exceedingly rare and aggressive neoplasm. Tunica vaginalis shares a common embryological origin with visceral pleura, peritoneum, and pericardium.[1,2] Malignant mesothelioma of the tunica vaginalis is morphologically identical to mesothelioma of the peritoneum,[1,2] however, only 0.3% to 5% of all malignant mesotheliomas arise from this origin.[2] The pathophysiology of these neoplasms remains unclear, and it is difficult to obtain a preoperative diagnosis. Historically, this neoplasm has been associated with a poor prognosis and there are no guidelines for management because of its rarity. Herein we report a contemporary case series of 7 patients who were diagnosed with malignant mesothelioma of the tunica vaginalis, our experience with early surgical management, and subsequent outcomes.

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Cases

Seven cases of histology confirmed malignant mesothelioma of the tunica vaginalis were identified between 2003 and 2014 on retrospective review of the National Institutes of Health (NIH) patient records. Clinical features, history of asbestos exposure, and primary therapeutic approach were collected and verified. Hematoxylin and eosin-stained slides for each case were reviewed with an expert pathologist and the histologic diagnosis of malignant mesothelioma was confirmed using established criteria. The tumors were then subclassified into epithelioid, or biphasic patterns on morphology. Immunohistochemically stains were reviewed. Follow-up data of patients were obtained by reviewing charts and corresponding with primary physicians.

Clinical Characteristics and Presentation

Clinicopathologic features of the series are summarized in Table 1. The mean age at diagnosis was 63.6 years (range, 43-85 years). Initial presentation included: 4 (57.1%) hydroceles, 1 (14.3%) solid scrotal mass, 1 (14.3%) with an inguinal mass mimicking an inguinal hernia, and 1 (14.3%) spermatocele. All

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Table 1 Summary of Clinical, Therapeutic, and Outcomes for Patients With Tunica Vaginalis Mesothelioma

Age, y	Ethnicity	Asbestos Exposure	Smoking	Presenting Syndrome	Histology	Immunohistochemistry Staining	Surgical Therapy	Adjuvant Therapy	Disease-Free Survival, mo
74	Caucasian	N	Y	Hydrocele	Biphasic	Positive for calretinin, WT1, cytokeratins A1/A3, and mesothelin (in 30% of tumor cells), and negative for CK5/6 and CEA	1. Hydrocelectomy 2. Left radical orchiectomy with excision of scrotal scar	None	Lost to FU
67	Caucasian	Y	N	Scrotal mass	Biphasic	Positive for AE1/AE3, EMA, calretinin, WT1, CK5, BER-EP4, D240 and focally positive for CD15. Negative for MOC-31, CEA, B72.3	1. Left radical orchiectomy	None	47
58	Caucasian	N	Y	Spermatocele	Epithelioid	Positive for calretinin, WT1, CK A1/A3, CK7, and mesothelin (2+ in 30% of tumor cells), and negative for CK5/6, CK20	1. Right inguinal orchiectomy	Radiation and chemotherapy	65
43	Caucasian	N	N	Scrotal mass	Epithelioid	NA	1. Right groin exploration inguinal herniorrhaphy 2. Right orchiectomy and hemiscrotectomy	None	12 Recurrences in peritoneum (14-month FU)
47	Caucasian	N	N	Hydrocele	NA	NA	1. Right orchiectomy 2. Left orchiectomy prophylactically	Radiation	155
85	Caucasian	Y	N	Hydrocele	Epithelioid	Positive for CK AE1/AE3, Calretinin and WT1. Negative for CK5/6. Mesothelin is positive, 2+ in 30% of tumor cells	1. Hydrocelectomy 2. Inguinal incision excision of scrotal scar and right orchiectomy	Radiation	19
71	Caucasian	N	N	Hydrocele	NA	Positive for CK5, CK7, CK20, CD31, CD34, calretinin, MOC-31, BER-EP4, vimentin, and WT1	1. Hydrocelectomy 2. Radical left orchiectomy	None	15

Abbreviations: AE1/AE3 = pan cytokeratin antibody; BER-EP4 = Ep-CAM/epithelial specific antigen; CD = cluster of differentiation; CEA = carcinoembryonic antigen; CK = cytokeratin; EMA = epithelial membrane antigen; FU = follow-up; MOC-31 = epithelial specific antigen/Ep-CAM; N = no; WT1 = Wilms tumor antibody; Y = yes.