Case Report

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Malignant mesothelioma of the *tunica vaginalis* testis: with comprehensive review of literature and case report

Anwar M Alesawi[1,2,3], Jerome Levesque[4] and Vincent Fradet[1,2]

Introduction

Malignant mesothelioma of the *tunica vaginalis* testis is a rare tumor. Since the first case report, fewer than 175 cases have been reported to-date.[1,2,3] Most of these cases presented with nonspecific symptoms, such as a painless scrotal mass associated with hydrocele. The pathogenesis of this malignant neoplasm is unclear and is mostly related to asbestos exposure,[4] as well as a long-lasting hydrocele.[5] An early preoperative diagnosis is difficult to obtain, and few cases have even been suspected to be mesothelioma.[3] We hereby report a case of malignant mesothelioma of the *tunica vaginalis* that was diagnosed preoperatively by scrotal hydrocelectomy.

Case report

A 69-year-old male patient was referred to our service with a right scrotal mesothelioma that was diagnosed incidently after having a recent right hydrocelectomy by finding mesothelioma elements in the submitted specimen.

There was no history of asbestos exposure or trauma to the testis.

Patient developed a progressing right scrotal mass three months after the hydrocelectomy, which was followed-up by ultrasonography and Doppler ultrasonography that confirmed the increase in the size of the mass.

Physically, patient was active, well, not in pain, sexually active and reported no lower urinary tract symptoms, but he had a solid, nodular mass at the scrotal surgical scar of previous surgery that was not associated with enlarged lymph nodes nor bone tenderness or pain. Ultrasonography (Figure 1) showed a hypoechogenic scrotal nodular mass, and Doppler ultrasonography (Figure 2) showed the nonvascular nature of this hypoechogenic mass. Testicular tumor markers were negative. Our surveillance time was almost one year before performing a right radical orchidectomy with hemiscrotectomy. The histopathology report confirmed the diagnosis of mesothelioma of *tunica vaginalis*.

Patient follow-up was one year after surgeryand showed no clinical signs of local or distant recurrence. Enhanced computed tomography was performed at three, six, nine and 12 months following surgery and showed no local recurrence or distant metastasis.

Macroscopic pathology

Around five lobulated nodules encircled the testis, the largest nodule measuring 3.0 × 2.5 × 2.0 cm. These nodular masses are whitish in colour, hard, partially gray and cystic.

These nodular lesions are well circumscribed and compress the testis (Figure 3).

Microscopic histopathology and immunohistochemistry

The tumor is partially tubulo-papillary and partially nonorganized solid nests that originate from the mesothelial layer in proximity to scrotal wall, infiltrating the testicle.

There is no sarcomatous differentiation.

The tumor strongly stained positive for mesothelin, calretinine and D240 and negative for Tag72, CEA and MOC31 (Figures 4–6).

[1]Urology Division, Université Laval, Canada
[2]Centre Hospitalier Universitaire de Québec Research Centre, Université Laval, Canada
[3]Ministry of Higher Education, Saudi Arabia
[4]Pathology Department, Université Laval, CHUQ-L'Hôtel-Dieu de Québec, Canada

Corresponding author:
Anwar M. Alesawi, 6, Jardins Merici, Apt 614, Quebec City, QC, G1S4N7, Canada.
Email: esawi_anwar@yahoo.com

Figure 1. Ultrasound image showing hypoechogenic mass.

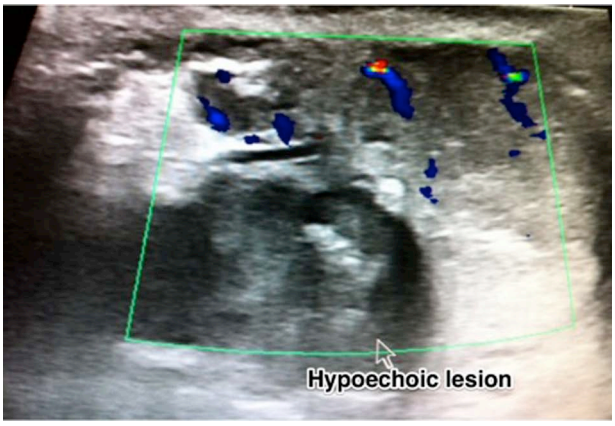

Figure 2. Doppler ultrasound image showing nonvascular lesion.

Figure 3. Mid-sagittal section of the testicle showing the whitish multinodular lesion compressing the testicular parenchyma.

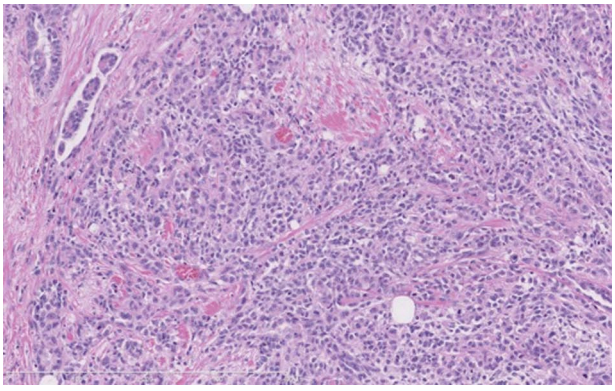

Figure 4. Tubulo-papillary pattern of mesothelioma (×40).

Discussion

Malignant mesothelioma of the *tunica vaginalis* testis is a rare tumor. We conducted a comprehensive review of the literature based both on a CD-Medline search of the literature and on the reference lists of published articles. Malignant mesothelioma was first described by Barbera and Rubino[6] in 1957; 53 such cases were found in the literature up to 1995 by Jones et al., 10 cases were found by Bisceglia et al. when he performed a literature review up to 2009, but we found 24 reported cases after the paper of Bisceglia et al. 54 not including our case, giving a total of 248 cases reported so far.

Malignant mesothelioma of the *tunica vaginalis* testis is present in 0.3%–5.0% of cases of malignant mesothelioma.[7,8] Mesothelioma in general is an uncommon malignancy arising from the epithelial membranes of the coelomic cavities, such as the pleura, pericardium and peritoneum. Mesotheliomas of the pleural and peritoneal cavity are relatively more common than scrotal tumors, which arise from the tunica, an outpouching of the abdominal peritoneum.

The tumor occurs most frequently in older men. Although the exact pathophysiology is unclear, contact with asbestos and a long-standing hydrocele are considered to be risk factors of malignant mesothelioma of the *tunica vaginalis*.[3,5,10] An association with asbestos exposure is present in 31%–41% of cases.[3,10] Asbestos is a heat-resistant material that exhibits superior durability and economy, and hence is still widely used in construction.

We conducted a comprehensive review of literature based both on a CD-Medline search of the literature and on the reference lists of published articles and we found 84 reported cases not including our case, 64 cases in which the patient presented at age 40 years or more. In our review, we found 16 cases that were associated with asbestos exposure, 42 cases without and 27 cases that were not specified (see Table 1).

In a review of nonasbestos-related malignant mesothelioma, it was found that chronic inflammatory processes

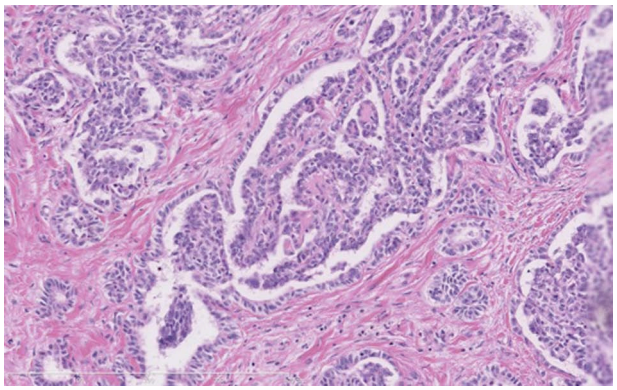

Figure 5. Nonorganized solid-nest pattern of mesothelioma (×40).

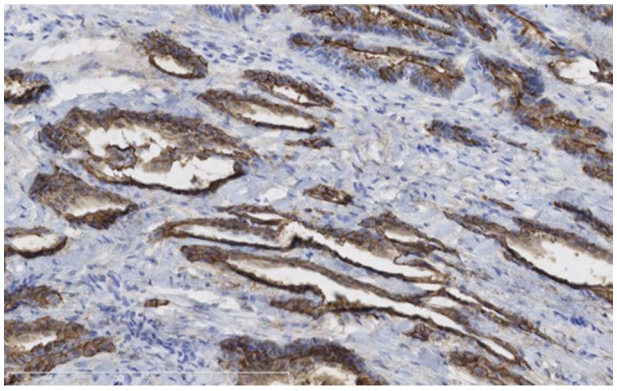

Figure 6. Immunohistochemistry for mesothelin, strongly positive (×20).

have also been suggested to be possible causes of malignant mesothelioma.[11]

A correct preoperative diagnosis is difficult and requires advanced imaging techniques, for example, ultrasound is reported to be useful in preoperative diagnosis,[12] while color Doppler ultrasound might have the potential to be used to unveil the vascular characteristics of malignant mesothelioma.[13,14]

Electron microscopy has been the gold standard for the diagnosis of mesothelioma. However, the International Mesothelioma Interest Group recommends that immunohistochemical examinations may replace the role of electron microscopy in identifying malignant mesothelioma, on account of their high sensitivity and specificity greater than 80% for suspicious lesions.[15] Tunica malignant mesothelioma has been reported to express cytokeratin 7, cytokeratin 5/6, epithelial membrane antigen, thrombomodulin and calretinin, but not cytokeratin 20, Ber-EP4,carcinoembryonic antigen or Leu-M1.[16]

Fine-needle aspiration (FNA) may prove to be an important and rapid diagnostic tool in such cases.[17] Considering the sensitivity of cytology and the potential risk of metastasis, the routine use of FNA for preoperative diagnosis is unclear.

Although this tumor is most often seen in patients between the ages of 55 and 75 years, 10% of patients are younger than 25 years,[3] with the main clinical symptom being painless scrotal mass with hydrocele.

In our review, we found 40/85 cases with hydrocele as a presenting symptom, 33/85 cases with scrotal mass and nine of 85 cases with scrotal pain as a presenting symptom (see Table 1).

Diagnosis is mostly made intraoperatively or post-operatively owing to the nonspecific symptoms and the absence of tumor markers before surgery, while commonly the preoperative impressions or diagnoses is hydrocele in 49.5% of cases, followed by the suspicion of a testicular tumor, which occurs in 36.6%[9] (see Table 1).

Treatment for malignant mesothelioma of the *tunica vaginalis* testis should start with surgery as first-line therapy in cases of early disease. Radical inguinal orchidectomy appears to be the optimum treatment. Local resection of the hydrocele wall is associated with a local recurrence rate of 36%, whereas local recurrence after orchidectomy is reported in 10.5% to 11.5% of patients.[3] Hemiscrotectomy is often required for local control mainly when the scrotal wall was violated, as when performing cytologies.

Retroperitoneal lymph node spread is found in 15% of cases, para-aortic lymph nodes are the first site of deposit, whereas pelvic (iliac and obturator) nodes are involved in more advanced stages.[2] Plas et al. recommended that if radiological examinations show no lymph node involvement, routine retroperitoneal lymph node dissection need not be performed because of the high negative rate.[3]

Adjuvant therapy with systemic chemotherapy and radiotherapy or both might provide a better overall survival rate in advanced disease.[18] Plas et al. reported the superiority of radiotherapy over chemotherapy as an adjuvant therapy.[3]

Malignant mesothelioma of the *tunica vaginalis testis* is an aggressive tumor with high recurrence and mortality rates; about 52% of patients develop local recurrence or metastasis within two years (see Table 1 for sites of recurrence by Plas et al.) and 40% of patients die from their disease, with a median survival of 24 months.[3,10] Patients younger than 60 years and organ-confined disease at diagnosis are significant factors correlated with survival as patients younger than 60 years have better survival and less aggressive clinical course than older patients.[3]

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

Table 1. Characteristics of reported mesothelioma cases

Pre-operative diagnosis	Number of cases	Percentage (%)	
Hydrocele	50	49.5	Yen et al. *World Journal of Surgical Oncology* 2012, 10:238. (Common pre-operative diagnosis of mesothelioma of the *tunica vaginalis* testis.)
Testicular tumor	37	36.6	
Inguinal hernia	6	5.9	
Epididymitis	3	3	
Spermatocele	2	2	
Testicular torsion	2	2	
Post-traumatic testicular lesion	1	1	
Total number	101	100	

(Development of metastasis in malignant mesothelioma of the *Tunica Vaginalis* Testis)		
Site of metastasis	% of patients	
Local recurrence	23.7 %	Plas E. et al: Malignant mesothelioma of the *tunica vaginalis* testis: review of the literature and assessment of prognostic parameters. *Cancer* 1998, 83:2437–2446
Lymph nodes	13.8 %	
Lung	9.7 %	
Liver	4.2 %	
Pleura	2.7 %	
Omentum	2.7 %	
Other (colon, spleen, mesentery, mediastinum)	5.6 %	

Presenting symptom	Number of cases	Age at presentation	Number of cases
Hydrocele	40	< 40 year	14
Scrotal mass	33	> or = 40 year	64
Scrotal pain	9	Unknown	7
Inguinal mass	3	Paratesticular mesothelioma. Review of the Literature cases (June 2002–September 2013) by Anwar Alesawi et al 2013.	
Epididymitis	3		
Incidental	1		
Unknown	11		

Asbestos exposure		
Yes	No	Unspecified
16	42	27

Conflict of interest

None declared.

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