

## Well-differentiated papillary mesothelioma of the tunica vaginalis: Case report and literature review



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### Introduction

Well-differentiated papillary mesothelioma (WDPM) of the tunica vaginalis testis originates from mesenchymal tissue; only 0.3%–5% of all cases of mesothelioma affect the tunica vaginalis.

WDPM may affect the spermatic cord, epididymis, and the peritoneal mesothelium of inguino-scrotal hernia sacs.<sup>1</sup> Compared with malignant mesothelioma (MM), WDPM appears usually not aggressive and accompanied by an indolent clinical behavior,<sup>2</sup> mimicking benign pathologies of the scrotum as hydrocele.

Here we present a case of WDPM of the tunica vaginalis testis and a literature review regarding the clinical and pathological features, diagnostic modalities, therapeutic aspects and prognosis of this rare entity.

#### Case report

A 83-year old man performed an urological consultation for a painless enlargement of the left scrotum for 12 months, which gradually progressed. Lower urinary tract symptoms and macrohematuria weren't detected. He presented dyslipidemia, hypertension and performed coronary angioplasty and stenting for ischemic cardiomyopathy; he hadn't a family history of genitourinary cancer. No history of significant asbestos exposure was found in this patient.

On physical examination, the left scrotum appeared enlarged with a size of 10 × 7 × 6 cm, without palpable mass. On ultrasonography a left hydrocele was detected with multiple iso-ipo-echoic masses, 1.3 cm

maximum diameter, located on the tunica vaginalis and albuginea testis (Fig. 1).

The patient underwent a left scrotal surgical exploration; straw-colored fluid yielded was found. Moreover, multiple lesions was located on the tunica vaginalis and albuginea of testis; some of these were excised to perform an immediate histopathological examination. The diagnosis of a WDPM of the tunica vaginalis was made. Under the circumstances of the multiple lesions and the patient's age, a radical left orchiectomy was performed. The patient had an uneventful recovery and was discharged on postoperative day 1. Specimen histopathological examination revealed multiple papillary lesions, soft and grey, from 0.1 to 1 cm, on the tunica vaginalis and albuginea testis. Testis, epididymo and spermatic cord were not involved. Immunohistochemical analysis showed no expression of K20; proliferation rate of Ki67 was 10%; calretinin, CK7 and vimentin were expressed (Fig. 2a–b). The diagnosis of a WDPM of the tunica vaginalis was confirmed, and the tumor appeared to have been completely excised.

As clinical staging, he performed a PET-CT scan revealed no pathological features. No further treatments were performed. After 6 months from surgery, the patient is alive and cancer free.

### Discussion

Mesothelial lesions of the paratesticular region include mesothelial cysts, reactive mesothelial hyperplasia, WDPM, and MM. Of these, WDPM is an uncommon tumor, usually detected in the peritoneum of women of reproductive age. It is infrequently seen in other anatomic

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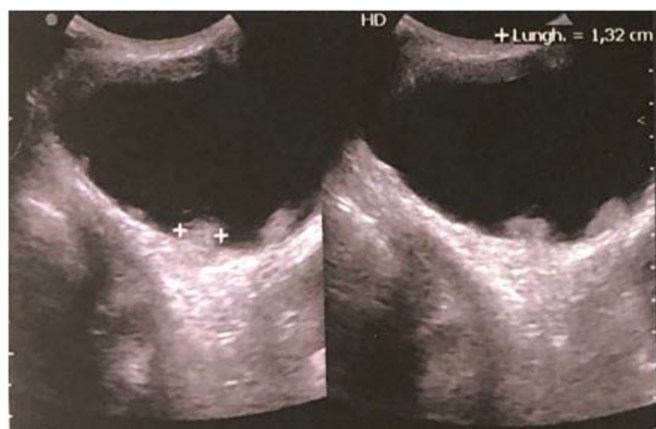


Fig. 1. WDPM ultrasonography aspect.

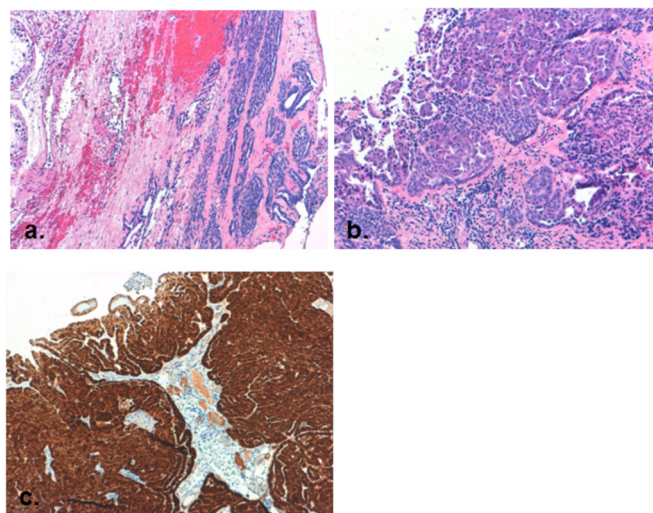


Fig. 2. Microscopic pathological features: a) Hematoxylin and eosin stain on tunica albuginea testis; b) Hematoxylin and eosin stain on tunica vaginalis testis; c) Expression of calretinin in immunohistochemical analysis.

sites, such as the pleura, pericardium, and tunica vaginalis. Only 0.3%–5% of all cases of mesothelioma affect the tunica vaginalis. WDPM of tunica vaginalis testis is rare and globally less than 20 cases have been reported in the literature.

The main differential diagnosis of WDPM includes mesothelial hyperplasia and MM. Mesothelial hyperplasia can also be composed of papillary structures, but often do not contain a fibrovascular core like WDPM.<sup>3</sup> MM present major features that should be considered in making a diagnosis as the presence of invasion, marked cytologic atypia, atypical mitosis, and a bulky mass.<sup>2</sup> Typically, WDPM should display morphological features of well-formed fibrovascular papillae, lined by a single row of cuboidal mesothelial cells with bland cytology and absent mitotic activity ( $\leq 1 \times 10$  HPF) without stromal infiltration or invasion of the adjacent tissues/organs. However, some WDPMs may show more complex pathologic features, such as areas of tubulopapillary differentiation and focal regions of solid sheets of cells; mesothelioma of tunica vaginalis of uncertain malignant potential (MUMP) has been proposed to describe lesions with more complex

tubulopapillary growth pattern.<sup>4</sup>

In literature, patients range in age from 18 to 70 years at presentation with a mean age of 43 years, but in the current case an age of 83 years was observed. Patients most commonly present with scrotal signs and symptoms as scrotal pain or swelling, but hydrocele is the most common presenting symptom as in the current case. On ultrasonography, as well as hydrocele, superficial small nodules on the tunica vaginalis testis, from a few millimeters to 3 cm size, are observed (as in the current case); this is the typically presentation of WDPM on ultrasonography.<sup>3</sup>

As clinical staging, is necessary to perform, as well as a scrotal ultrasonography, a stadation PET-CT, under the circumstances that differential diagnosis of WDPM includes MM and MUMP, to achieve the absence of pathological features.

The etiology or tumorigenesis of WDPM of the tunica vaginalis remains unknown. It has been suggested that some papillary proliferative lesions might be related to local trauma and inflammation. However, in our case there was no evidence of such insults, and no presence of chronic inflammation was found in the lesion itself. For well-studied malignant mesotheliomas of pleura, in addition to prolonged exposure to asbestos, infection by SV40 virus has recently been indicated etiologically.<sup>5</sup> No history of significant asbestos exposure was found in this patient.

Appropriate treatment for WDPM appears to be the complete surgical excision, which may be a radical orchiectomy in cases as present case where multiple lesions are presents. Given that there have been no observed cases of metastasis upon diagnosis or on follow-up of all cases of WDPM reported, WDPM is assumed to be a benign tumor.

At present, there is no recommended follow-up for WDPM. Some authors recommend a close and conservative follow-up regime similar to that of malignant mesothelioma, some other ones consider it may be excessive.

WDPM is not well-known because of such a limited number of cases. A large series is needed to answer the questions about this enigmatic tumor.

## Conclusion

WDPM of the tunica vaginalis testis is a rare entity. It is important to have a differential diagnosis compared to MM, given that WDPM appears usually not aggressive and accompanied by an indolent clinical behavior. Appropriate treatment for WDPM appears to be the complete surgical excision. WDPM is assumed to be a benign tumor, given that the absence of reported cases of metastasis.

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