

## Oncology

## Bilateral Malignant Mesothelioma of Tunica Vaginalis A Case Report on Rare Presentation



Ashhad A. Shaikh\*, Kishor V. Naik, Shisir N. Shetty, Noor N. Ansari, Pallavi S. Babhale

Naik Nursing Home &amp; Endoscopy Centre, Sector 12 E, Balaji Garden, Koparkhairane, Navi Mumbai, 400709, Maharashtra, India

## ARTICLE INFO

## Article history:

Received 2 May 2017

Accepted 7 June 2017

## Keywords:

Malignant mesothelioma

Bilateral

Tunica vaginalis

Asbestos

Calretinin

## ABSTRACT

Malignant mesothelioma involving the para-testicular tunica is extremely rare and an aggressive tumor. Bilateral malignant mesothelioma of the tunica vaginalis is not reported previously in the literature. Rarity of the disease, absence of any specific clinical and radiological findings makes the preoperative diagnosis difficult. Aggressive surgical approach is the key to successful management with high inguinal orchiectomy and scrotectomy appears to be optimal treatment in patients with localized disease.

© 2017 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## Introduction

Malignant paratesticular mesothelioma is usually related to asbestos exposure while cases unrelated to asbestos exposure are also reported.<sup>1</sup> Synchronous bilateral paratesticular mesothelioma are very rare. We came across a case of bilateral malignant mesothelioma of the tunica vaginalis presented as scrotal swellings in a 65-year-old male with no known exposure to asbestos.

## Case report

A 65-year-old male presented with bilateral painless scrotal swellings more on the right side with heaviness and dragging sensation gradually increased over six months. Past medical, surgical and family history were non-contributory. Environmental and professional exposure to asbestos was not present. Clinical examination revealed bilateral scrotal swelling with right measuring 70 × 50 × 50 mm, 45 × 40 × 35 mm on the left non-tender on palpation. No hydrocoele, thickening of epididymis or spermatic cord was evident on the clinical examination. Ultrasound of right and left testes showed heterogeneous, lobulated, hypoechoic mass lesion with variable vascularity arising from the tunica vaginalis and compressing and infiltrating the testicular parenchyma. Testicular tumor markers including  $\alpha$  fetoprotein,  $\beta$  sub-set of human

chorionic gonadotropin were within normal limit. High inguinal orchiectomy with hemiscrotectomy was performed on the right side as consent for simultaneous bilateral orchiectomy was deferred.

Gross examination revealed tumor infiltrating the scrotal skin with diffuse concentric thickening of the tunica by grayish-white fleshy vaguely lobulated mass compressing the testicular parenchyma (Fig. 1) encasing and infiltrating of the adjacent structures.

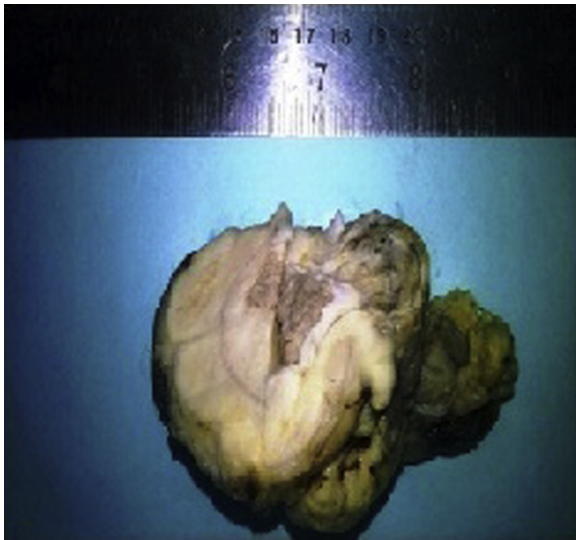
Histological sections revealed biphasic tumor comprising admixture of epithelial and sarcomatous element infiltrating the atrophic testicular parenchyma. Epithelial component were characterized by arrangement of mesothelial cells in tubules, papillae and sheets with neoplastic cells showing high nucleo-cytoplasmic ratio, prominent nucleoli and eosinophilic cytoplasm (Fig. 2). Psammoma bodies were not seen. Sarcomatous element comprised bizarre looking stromal cell arranged in sheets.

Tumor cells showed diffuse reactivity to calretinin, (Fig. 3), cytokeratin 5, 6, 7 vimentin and epithelial membrane antigen. Carcino-embryonic antigen, estrogen and progesterone receptors were negative confirming a diagnosis of malignant biphasic mesothelioma of the tunica vaginalis.

After explaining prognostic implications of the histopathology report consent was obtained and left high inguinal orchiectomy with hemiscrotectomy were performed. Gross pathology revealed thickening of the tunica with asymmetric infiltration in the testicular parenchyma by fleshy mass and similar histological and immunohistochemical profile as on right. Subsequent positron emission tomography revealed no residual disease. Post-operative

\* Corresponding author.

E-mail address: [histosolutions@gmail.com](mailto:histosolutions@gmail.com) (A.A. Shaikh).



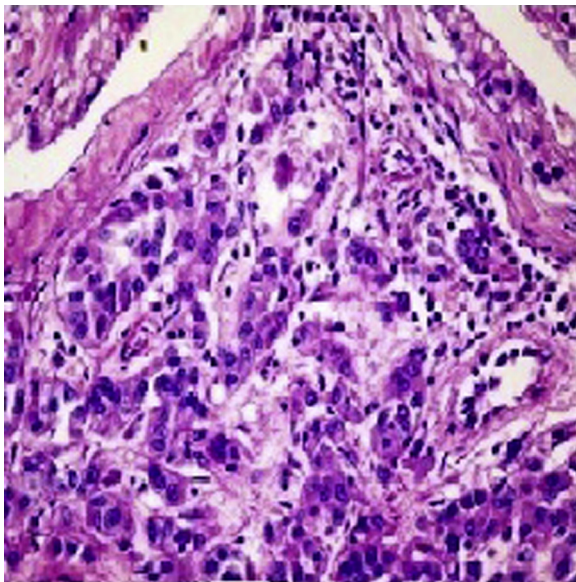
**Figure 1.** Gross specimen showing lobulated mass involving the tunica vaginalis.

stay was uneventful. As chemotherapy and radiotherapy plays no proven role in management in patients with localized disease our patient had consented to close observation. Positron emission tomography studies were negative for local recurrence or distant metastases on completion of 24 month.

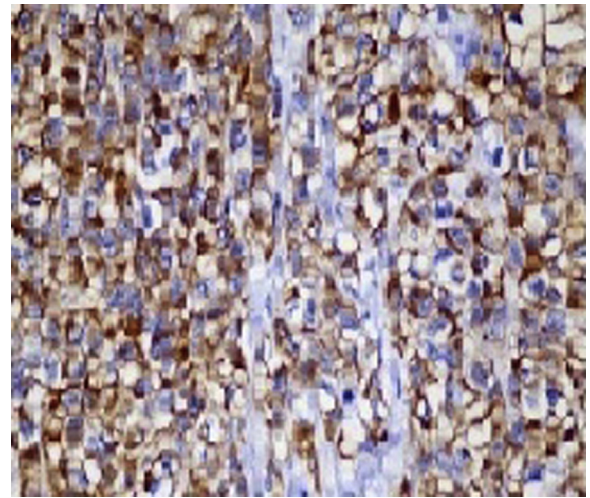
### Discussion

Involvement of the para-testicular tunica vaginalis by mesothelioma is reported in about 0.3–1.4% of all malignant mesotheliomas. We came across only one case of synchronous paratesticular mesothelioma in literature. Though cases has been reported in age group of 18–77 years majority presented in the 6th to 8th decade.<sup>1</sup> Age at diagnosis in our patient was 65 year.

In a review of non-asbestos –related malignant mesothelioma, chronic inflammatory processes have been suggested to be possible causative factor.<sup>1</sup> Exposure to fluoroedenite, ionizing radiation with



**Figure 2.** Hemotoxylin and Eosin showing tubulo-papillary pattern.



**Figure 3.** Immunohistochemistry showing diffuse reactivity to calretinin.

use of thoratrast along with genetic component have also been implicated in the genesis of mesothelioma.

Painless scrotal mass with hydrocoele is the commonest presentation of malignant mesothelioma of the tunica vaginalis.<sup>2</sup> Due to non specific presentation, clinical and radiological differential usually includes hydrocoele, testicular tumor, inguinal hernia and epididymitis.<sup>3</sup> Patient in discussion presented with painless gradually increasing bilateral scrotal swelling with dragging sensation over six months.

Ultrasound suggested as most popular aid in preoperative evaluation of scrotal swellings commonly show hydrocoele, extra testicular mass with atypical features of variable vascularity on color Doppler<sup>3</sup> Computed tomography is helpful in staging of tumor, diagnose local and distant metastases including evaluation of retroperitoneal lymph nodes. Aspiration cytology from the hydrocoele fluid has been described in few cases of rapidly growing hydrocoele in elderly however considering the low sensitivity and potential risk of metastasis, routine use of fine needle aspiration cytology is still not established. Clinical utility of serum hyaluronic acid as prognostic marker is still under evaluation.

Variable histological patterns including epithelioid, tubo-papillary and solid architectural types are described. Few cases showing biphasic pattern with variable epithelial and malignant stromal element has also been described as was seen in our case.

Immuno-histo-chemical positivity for calretinin, vimentin, Wilms tumor-1, epithelial membrane antigen, D2-40, thrombomodulin, cytokeratin 7 and cytokeratin 5/6 had been documented. In our case tumor cells showed diffuse reactivity to calretinin, cytokeratin 5,6,7, vimentin, and epithelial membrane antigen.

Protocols for the treatment of malignant mesothelioma of the tunica vaginalis are not established due to extremely low incidence and difficult pre-operative diagnosis. Being an aggressive tumor with high recurrence rate of 36% after local resection of hydrocoele wall, hemiscrotectomy is often required for control of disease. Local recurrence after orchiectomy is 10.5–11.5%.<sup>4</sup> Primary surgical management has been the treatment of choice with high inguinal orchiectomy with hemiscrotectomy appears to be optimum treatment as was performed in our case. Median survival has been reported as 24 months.<sup>4</sup>

Open retroperitoneal lymph node dissection and use of systemic chemotherapy and radiotherapy is recommended in advanced

cases for overall improvement in the survival in line of diffuse peritoneal mesothelioma.<sup>5</sup> High inguinal orchidectomy with hemiscrotectomy was offered as primary treatment with active surveillance and close follow-up. Patient remains disease-free at twenty 24month post operative.

### Conclusion

As the etiopathogenesis of paratesticular malignant mesotheliomas is still obscure it should be considered in the differential diagnosis of extratesticular tumor in the scrotum with normal tumor marker profile irrespective of exposure to asbestos and other predisposing factors. Bilateral involvement, biphasic histology, prompt diagnosis and early surgical approach with 24months disease free follow-up make our case for publication. We therefore recommend an early and aggressive surgical approach for successful management of disease.

### Conflict of interest

The authors declare that they have no conflict of interests.

### References

1. Garcia de Jalon A, Gil P, Azua-Romeo J, Borque A, Sancho C, Rioja LA. Malignant mesothelioma of the tunica vaginalis. Report of case without risk factors and review of literature. *Int Urol Nephrol.* 2003;35:59–62.
2. Jones MA, Young RH, Scully RE. Malignant mesothelioma of the tunica vaginalis. A clinicopathologic analysis of 11 cases with review of literature. *Am J Surg Pathol.* 1995;19:815–825.
3. Fields JM, Russell SA, Andrew SM. Ultrasound appearances of a malignant mesothelioma of the tunica vaginalis testis. *Clin Radiol.* 1992;128–130.
4. Plas E, Riedl CR, Pfluger H. Malignant mesothelioma of the tunica vaginalis testis; review of literature and assessment of prognostic parameters. *Cancer.* 1998;83(12):2437–2446.
5. Yan TD, Welch L, Black D, Sugarbaker PH. A systemic review on the efficacy of cytoreductive surgery with perioperative intraperitoneal chemotherapy for diffuse malignant peritoneal mesothelioma. *Ann Oncol.* 2007;18:827–834.