CASE REPORTS

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Atypical presentation of testicular mesothelioma



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Abstract

Mesothelioma is a rare cancer that generally affects the lining of the lungs. However, in more rare cases, there are extra-pleural involvement of organs that are lined with mesothelial cells, including the testicle. Mesothelioma of the tunica vaginalis of the testicle typically presents with a painless scrotal mass with accompanying hydrocele of the affected side. The most common predisposition to mesothelioma of the tunica vaginalis is previous asbestos exposure, long-standing hydrocele, and previous scrotal surgeries. In this case we present a patient with isolated left tunica vaginalis mesothelioma of the testicle, yet without classic risk factors or other sites of disease. Left radical orchiectomy was performed and subsequent follow-up imaging did not reveal any sites of recurrence or metastasis. Although, mesothelioma of the tunica vaginalis is a rare cancer, it should be seen as a possible differential when evaluating new testicular masses.

Keywords Mesothelioma, Tunica vaginalis, Hydrocele, Orchiectomy, Testicular tumor

1 Background

Mesotheliomas are rare tumors that generally arise after exposure to asbestos and often present in the pleura or peritoneum. However, more rarely, mesotheliomas can occur in extra-pleural tissue that is lined by mesothelial cells, such as the peritoneum and the tunica vaginalis. Mesotheliomas arising from the tunica vaginalis are rare with less than 300 documented cases and comprising less than 5% of all mesothelioma tumors [1]. Asbestos exposure is a primary risk factor for the development of mesothelioma of the tunica vaginalis, though inflammatory causes such as trauma and long-standing hydrocele are also risk factors [1, 2]. These tumors most commonly arise in middle-aged men with a mean age of 53.5 years. The clinical presentation is typically of insidious onset with unilateral painless enlargement of the scrotum with

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accompanying hydrocele. An important ultrasonic finding is thickening of the tunica vaginalis [3]. The overall survival in patients with mesothelioma of the tunica vaginalis is 1.5 years with a 5 year overall survival of 49% [4]. In this case report we describe a case of malignant mesothelioma without the common presentation or associated risk factors.

2 Case report

A 60-year-old male presented to a urology clinic with a left scrotal lump that had been present for "several years" but was painless and stable in size. A scrotal ultrasound was ordered and displayed a 1.5×1.0 cm solid mass arising from the left epididymis (Fig. 1). Serum tumor markers, alpha fetoprotein (AFP), lactate dehydrogenase (LDH), and beta human chorionic gonadotropin (β -HCG), were ordered, and all of which were negative. A left radical orchiectomy was then performed. The patient's pathology from the radical left orchiectomy revealed malignant mesothelioma of the tunica vaginalis (Fig. 2). The tumor was measured at 3.0cm in size and all margins were negative. The patient was then sent for a computed tomography (CT) of the chest, abdomen and



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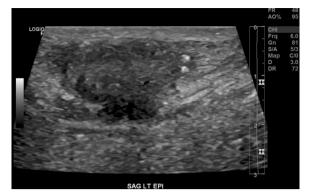


Fig. 1 Ultrasound Image of a 1.5 × 1.0 cm mass within the left epididymis

pelvis. The CT scan did not reveal any local adenopathy or evidence of metastatic disease. He had a repeat CT scan at 1 and 2 years postoperatively, and is currently on a surveillance protocol without any recurrence of disease after 2 years.

3 Discussion

Mesotheliomas stem from mesothelial cells which make up the lining of the pericardium, pleura, lungs, and testicles. Mostly commonly, mesotheliomas arise from exposure to asbestos. Asbestos induces DNA damage via creation of radical oxygen and nitrogen species, and iron free radicals. These in turn can cause DNA substitutions, deletions, and insertions. In addition to the creation of DNA damage, asbestos exposed mesothelial cells continually proliferate via the NF-kB pathway [5].

In addition to asbestos exposure, chronic inflammation can induce mesothelial cells to undergo oncogenic transformation. Cases of pleural mesothelioma have been documented without asbestos exposure but were subject to recurrent lung infections, pulmonary tuberculosis, and recurrent pleural effusions. For testicular mesotheliomas, it is posited that long-standing hydroceles and persistent inflammation in the tunica vaginalis predispose individuals to mesothelioma [6].

When approaching a scrotal mass, a thorough history and physical exam is required. It is important to inquire about a family history of testicular cancer, previous inguinal hernia repair, cryptorchidism, and previous orchiopexy. When physical exam confirms a solid mass, trans-scrotal ultrasound is the primary imaging technique to further investigate the solid mass. Seminomas are the most common testicular carcinoma in men older than 35 and it is imperative to retrieve serum tumor markers, including α FP, LDH, and β -HCG [6–8]. In contrast to testicular masses, solid epididymal masses,

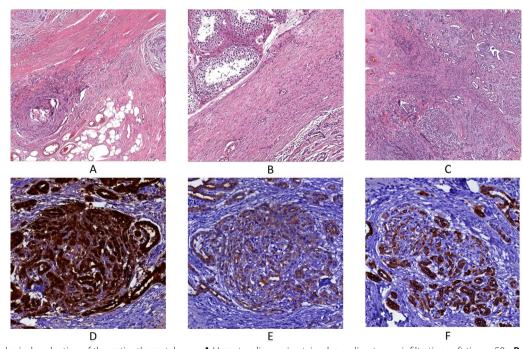


Fig. 2 Histological evaluation of the patient's scrotal mass. **A** Hematoxylin–eosin-stained revealing tumor infiltrating soft tissue, 50x. **B** Hematoxylin–eosin-stained showing fibrosis and tubular proliferation with normal spermatogenesis occurring, 100x. **C** Hematoxylin–eosin-stained demonstrating tubular and papillary mesothelial proliferation, 50x. **D** Immunohistochemical staining for Calretinin positive, 200x. **E** Immunohistochemical staining for D2-40 positive, 200x. **F** Immunohistochemical staining for CK5/6 positive, 200x

however, are typically benign. The incidence of malignancy is correlated with epididymal mass size greater than 1.5 cm and is generally regarded as the cutoff for intervention [9].

Mesothelioma of the tunica vaginalis is a rare form of an already uncommon cancer. The majority of reported cases have been found incidentally during hydrocelectomies [10]. The presentation in this case report was atypical. Commonly patients present with risk factors for mesothelioma of the tunica vaginalis. Our patient presented with a long-standing painless testicular mass, lack of primary asbestos exposure and lack of a concomitant long-standing hydrocele. Moreover, he had no other sites of mesothelioma, indicating a primary mesothelioma of the tunica vaginalis of the testicle.

In conclusion, mesothelioma of the tunica vaginalis is a rare form of an already rare cancer. Despite having notable risk factors and found in conjunction with longstanding hydrocele it should be kept on the differential diagnosis when evaluating a middle-aged male presenting with a testicular mass.

Abbreviations

β-HCG	Beta human chorionic gonadotropin
LDH	Lactate dehydrogenase
AFP	Alpha fetoprotein
PSA	Prostate specific antigen

Acknowledgements

Not applicable.

Author contributions

SR made significant contributions researching and writing this report. SN made significant contributions performing research for this article. NF was responsible for treatment plan and performance. NF also made a significant critical assessment of this article and edits for this report. All authors agree on the final version of the manuscript.

Funding

All the authors have no funding sources to declare relevant to this report.

Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate Not applicable.

Consent for publication

Written informed consent for publication of case report was obtained from the participant patient.

Competing interests

There was no funding for the study and no conflicts of interest to disclose.

Received: 20 December 2022 Accepted: 30 November 2023 Published online: 17 December 2023

References

- Goel A, Agrawal A, Gupta R, Hari S, Dey AB (2008) Malignant mesothelioma of the tunica vaginalis of the testis without exposure to asbestos. Cases J 1(1):310. https://doi.org/10.1186/1757-1626-1-310.PMID:19014 568;PMCID:PMC2596792
- Akin Y, Bassorgun I, Basara I, Yucel S (2015) Malignant mesothelioma of tunica vaginalis: an extremely rare case presenting without risk factors. Singapore Med J 56(3):e53–e55. https://doi.org/10.11622/smedj.2015053. PMID:25820862;PMCID:PMC4371211
- Chekol SS, Sun C-C (2012) Malignant mesothelioma of the tunica vaginalis testis: diagnostic studies and differential diagnosis. Arch Pathol Lab Med 136(1):113–117
- Nazemi A, Nassiri N, Pearce S, Daneshmand S (2019) Testicular mesothelioma: an analysis of epidemiology, patient outcomes, and prognostic factors. Urology 126:140–144. https://doi.org/10.1016/j.urology.2019.01. 009. (Epub 2019 Jan 19 PMID: 30668959)
- Yang H, Testa JR, Carbone M (2008) Mesothelioma epidemiology, carcinogenesis, and pathogenesis. Curr Treat Options Oncol 9(2–3):147–157. https://doi.org/10.1007/s11864-008-0067-z
- Yen CH, Lee CT, Su CJ et al (2012) Malignant mesothelioma of the tunica vaginalis testis: a malignancy associated with recurrent epididymitis? World J Surg Onc 10:238. https://doi.org/10.1186/1477-7819-10-238
- Ghazarian AA, Rusner C, Trabert B, Braunlin M, McGlynn KA, Stang A (2018) Testicular cancer among US men aged 50 years and older. Cancer Epidemiol 55:68–72. https://doi.org/10.1016/j.canep.2018.05.007.PMID: 29807233;PMCID:PMC6668029
- Gaddam SJ. Testicle cancer [Internet]. StatPearls [Internet]. U.S. National Library of Medicine; 2021 [cited 2021Oct19]. Available from: https://www. ncbi.nlm.nih.gov/books/NBK563159/
- Alleman WG, Gorman B, King BF, Larson DR, Cheville JC, Nehra A (2008) Benign and malignant epididymal masses evaluated with scrotal sonography: clinical and pathologic review of 85 patients. J Ultrasound Med 27(8):1195–1202. https://doi.org/10.7863/jum.2008.27.8.1195. (PMID: 18645078)
- Drevinskaite M, Patasius A, Kevlicius L, Mickys U, Smailyte G (2020) Malignant mesothelioma of the tunica vaginalis testis: a rare case and review of literature. BMC Cancer 20(1):1–6

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