Case Report

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Surgery Section

A Rare Case of Bilateral Paratesticular Malignant Mesothelioma of Tunica Vaginalis with Bilateral Hydrocele

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ABSTRACT

Paratesticular region is an indeterminate anatomical region including the spermatic cord with its contents- tunica vaginalis of the testes, epididymis and its appendices and testis. Paratesticular tumours may mimic testicular tumours and is clinically indistinguishable. Paratesticular malignant mesothelioma is an extremely rare tumour, representing only 0.3-5% of all mesotheliomas. There is not a specific clinical presentation however, it may present as a mass which is painless or as a hydrocele, having a wide age distribution. Lack of specific tumour markers presents a challenge in its diagnosis and management. Extreme diagnostic difficulty arises preoperatively in distinguishing malignant and benign tumour. Most of the cases are diagnosed intraoperatively or postoperatively when found on biopsy. Pleural and peritoneal mesotheliomas are known to occur in individuals exposed to asbestos. The case reported here is of a 32-year-old male who reported in the Out Patient Department (OPD) with the complaint of painless bilateral enlarged scrotum without history of exposure to asbestos. Bilateral orchidectomy followed by biopsy reported paratesticular malignant mesothelioma. Bilateral malignant mesothelioma of tunica vaginalis with bilateral hydrocele occurrence is rare, and it has an aggressive clinical progression, so clinical presentation and histopathology features are being presented to emphasise on the fact that it is critical for the urologists to be aware of such potentially rare entity. A mandatory high index of clinical suspicion is required, as lack of knowledge would result in an unnecessary radical orchidectomy as happened in present case.

Keywords: Malignancy, Mesothelial tumour, Orchiectomy, Testis

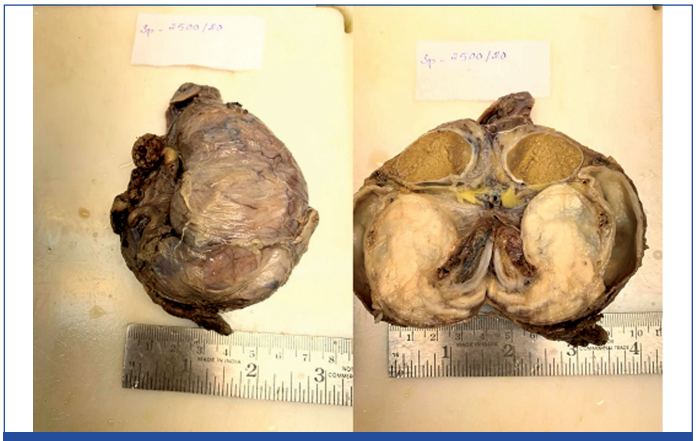
CASE REPORT

A 32-year-old male presented with bilateral scrotal swelling over six months associated with loss of weight. History of exposure to asbestos was not present. However, patient had no complains of pain and fever or any other significant co-morbidity. Medical, surgical and family history were not significant.

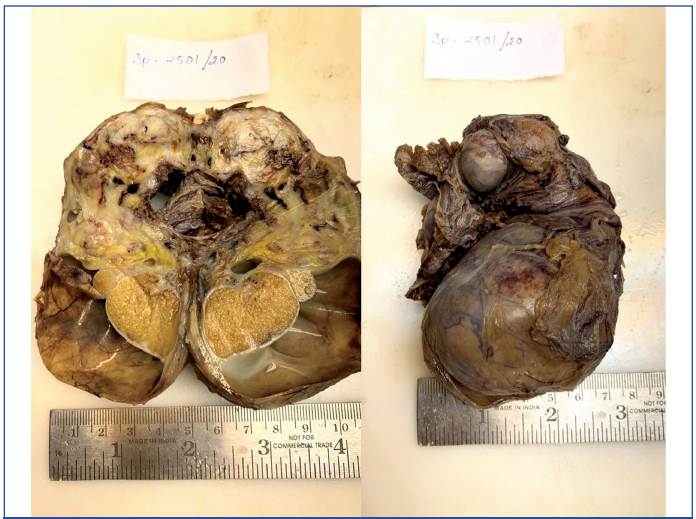
Physical examination revealed bilateral testicular swelling without hernia or any lymph node enlargement in inguinal region. Ultrasound (USG) revealed bilateral heterogeneous paratesticular mass of size-right -4×3.5×3 cm and left -6×4×2.5 cm with bilateral mild hydrocele. Computed Tomography (CT) abdomen was done which showed no distant lymph node involvement. Testicular tumour markers including α-feto-protein, β-HCG (Human Chorionic Gonadotropin) were within normal limit.

Considering it as malignancy arising from testis and infiltrating the paratesticular tissue, bilateral high inguinal orchidectomy was done after two days of hospitalisation. Gross examination revealed greyish white lobulated mass measuring right-sided -4.6×3.6×2.8 cm and left-sided -5.8×3.1×2.6 cm compressing both testes and not infiltrating the adjacent structures. [Table/Fig-1,2]. The cord was free of any malignancy on gross examination. Histopathology revealed tumour comprising of epithelial element arranged in tubulopapillary pattern [Table/Fig-3]. Psammoma bodies were not seen.

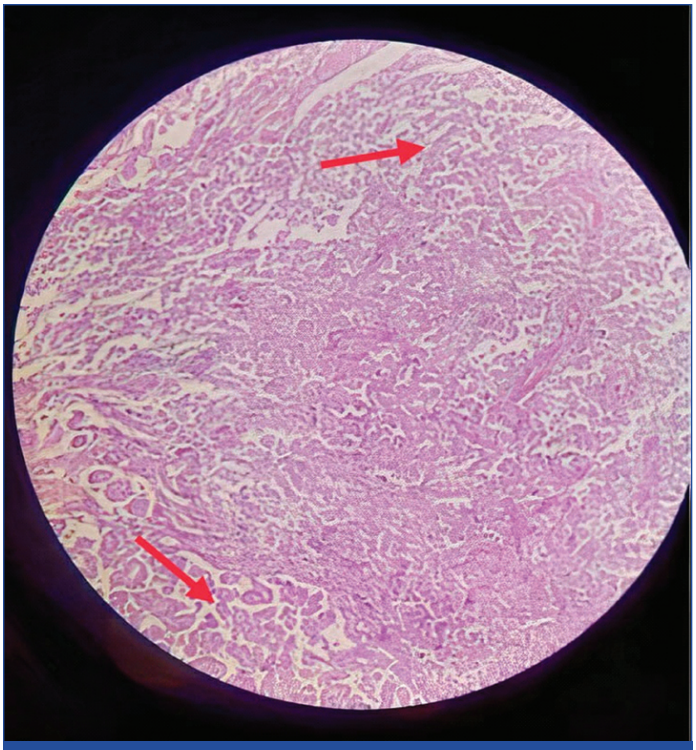
Oestrogen and progesterone receptors, carcino-embryonic antigen, were negative making a diagnosis of Stage I malignant mesothelioma. The patient was referred to oncologist for further management post-surgery with an advice to follow-up. After three months, following referral to oncology centre, patient came for follow-up with complains of painful swellings in both the groin suggesting metastatic nodes. On examination, lymph nodes in the bilateral groin were enlarged. CT scan was done and it showed multiple enlarged lymph nodes in bilateral groin, para-aortic region and the posterior mediastinum. Patient was started on chemotherapy by the oncologist which included 2 cycles of Pemetrexed and Cisplatin. After 30 days,



[Table/Fig-1]: Gross appearance and cut open right orchiectomy specimen shows nodular masses encasing the compressed testicular tissue.



[Table/Fig-2]: Cut open left orchiectomy and resected specimen shows nodular masses encasing the compressed testicular tissue.



[Table/Fig-3]: Photomicrographs of tumour cells showing tubulopapillary structures (arrows) Haematoxylin and Eosin, 40x.

there was progressive decline in the patient's performance status indicating disease progression. Considering his condition, he was sent for palliative care but died after three weeks.

DISCUSSION

Malignant mesotheliomas are rare tumours, which develops from the lining epithelium of coelomic cavities of the body. Mesothelioma of the lining of tunica vaginalis of testis represent only 0.3-5% of all mesothelial neoplasms [1]. The incidence rate is 18.6 times higher above 80 years than under the age of 50, suggesting increasing incidence with age [2]. Exposure to asbestos is a known risk factor for the development of pleural as well as peritoneal mesotheliomas but its link to tunica vaginalis tumours is not well established [1,2].

In the literature review done by Mezei et al., among the 89 reported cases of testicular mesotheliomas the possibility of asbestos exposure was considered for 50 patients (56%) and confirmed or assumed for 30% [2]. Most common age group affected by testicular mesothelioma are between 55 and 75 years [3]. Developmental mechanism of testicular mesothelioma remains poorly understood, risk factors include long term hydrocele, trauma and prior hernia surgery in the form of herniorrhaphy [4-6]

Hatzinger M et al., noted the literature has more than 80 reported cases of mesothelioma of testis [7]. The present case was a 32-year-old patient, with no risk factor and known history of asbestos exposure is being reported for addition to this list. Similar to this case, patients with testicular mesothelioma may present with scrotal problems [3]. USG is non invasive and has 90% accuracy in detecting testicular tumours [8]. USG can be used to exclude other conditions like epididymitis; adenomatoid tumour; hyperplasia of mesothelium; carcinoma of rete testis; serous type of papillary tumour; pleomorphic sarcomas; and various types of testicular germ cell tumours. The levels of tumour markers like αFP, β-HCG, and LDH can help in the diagnosis of tumours arising from testis [8]. However, the levels were normal in present case.

Seminomatous testicular tumour may have normal levels of tumour markers, as in index case, and an intraparenchymal mass may be seen by USG in seminoma patients [9]. In present case, bilateral heterogenous paratesticular masses were detected but both the testicular parenchyma was normal. The patient history and findings of the USG suggested an uncommon paratesticular region malignancy. In this situation, CT should be done for staging, localising distant metastasis, and involvement of retroperitoneal lymph nodes [10].

No distant metastasis was found in present case. Clinical evaluation, and pathological findings are important for differential diagnostic considerations including the various types of germ cell tumours particularly with involvement of testicular parenchyma (seminomas, embryonal carcinomas, and intratubular germ cell tumours), florid mesothelial hyperplasia of mesothelium, adenomatoid tumour, serous papillary tumours and rete testis carcinoma. When dealing with the biphasic variant, pleomorphic sarcomas should be considered. The prognosis remains grave, with a median survival of 23 months. To aid in accurate diagnosis immunohistochemical reactivity to calretinin, cytokeratin 5/6, and Wilms' tumour gene 1 should be done [11]. Not done in present case due to unavailability.

Treatment of testicular mesothelioma may include surgery, radiation therapy, chemotherapy and combination therapy in the higher stages [12]. For localised disease, as in present case, radical surgery in the form of inguinal orchidectomy should be offered as a surgical approach. Inguinal lymph node dissection is required if any evidence of metastasis in inguinal lymph nodes. Because of limited number of cases reported, benefit of offering adjuvant chemotherapy and/or radiotherapy is not clear. Chemotherapeutic agents like Cisplatin and pemetrexed can be offered to patients [13], while to prevent disease recurrence, radiotherapy may be of help [7]. Patient was surgically treated at an early stage and did not receive radiotherapy as no sign of metastasis but was offered chemotherapy due to progression of the disease post-surgery. Over a mean follow-up period of two years, 53% mortality has been reported in patients of testicular mesotheliomas [6].

Present case differs from those in the literature as it was without any risk factors for mesothelioma of testis, there was lack of specific clinical and radiologic features, and histological diversity. The prognosis remains poor regardless of adjuvant therapies based on extra testicular mesothelioma [14].

CONCLUSION(S)

The case reported here is a rare entity as, without exposure to known risk factors, but the clinical presentation and age fit the cases reported in literature. Apparently, a local disease but with fast progression and metastatic potential. A high index of clinical suspicion is required in making an accurate diagnosis of this rare entity. This tumour should be kept in mind as a differential diagnosis of mass of scrotal region, even at first it appears to be a hydrocele, regardless of asbestos exposure is there or not.

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