Malignant Mesothelioma of the Tunica Vaginalis Testis: A Rare Malignant Tumor without Asbestos Exposure

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Abstract:
Malignant mesothelioma of the tunica vaginalis testis is an extremely rare and often a deadly tumor. Preoperative diagnosis is very difficult due to the lack of specific clinical manifestations, mainly including hydrocele formation and painless inguinal mass. In this abstract, we presented a case of a young patient with hydrocele and dull pain of left testes, who was planned for laparoscopic repair of left inguinal hernia, during which a mass attached to the scrotal cord was discovered. Dissection and release of the mass from the cord structure were performed, and postoperative pathological examination revealed malignant mesothelioma arising from the left tunica vaginalis testis. The case was then referred to an oncology center where intraperitoneal hyperthermic chemotherapy with cytoreductive surgery was performed.

Keywords: Malignant mesothelioma, Tunica vaginalis, Mesothelioma, Testicular mass, Medical history, Tumor.

1. BACKGROUND
Malignant mesothelioma is a very rare tumor that is associated with multiple risk factors but can occur in the absence of any risk factor, the presentation usually lacks specific clinical manifestations and thus the preoperative diagnosis is very challenging and unusual.

2. INTRODUCTION
Mesotheliomas are relatively uncommon tumors that originate from serosal surfaces of pleura, peritoneum and pericardium. They usually occur after asbestoses exposure for several years in the past, other possible predisposing risk factors include trauma, long term hydrocele and herniorrhaphy [1], but the disease may also affect patients with no obvious risk factors. Rarely, they arise from the tunica vaginalis testis and this represents 0.3% to 5% of all malignant mesotheliomas [2]. However, testicular mesothelioma is often a deadly tumor that originates from mesenchymal tissue, formed from the evagination of the abdominal peritoneum into the scrotum and usually diagnosed postoperatively due to its rarity and unspecific clinical presentation.

3. CASE REPORT
A 27-year-Emirates, college graduate man, with no previous medical history, was previously diagnosed with left hydrocele, visited the Department of General Surgery of Al Qassimi Hospital on October 2018, due to dragging pain in the left inguinal region and mild enlargement of the left scrotum for 6 months, which gradually progressed. On physical examination, the left scrotum appeared to be normal with no palpable mass. The patient had positive cough impulses in the left groin, and suspicion of left inguinal hernia raised. Routine hematologic and biochemical parameters were within normal limits, ultrasonography revealed left cord hematoma like mass; query hydrocele, (Figs. 1a-1d). The plan was to do laparoscopic intraperitoneal hernia repair.

Operative report: In November 2018, laparoscopic intervention with insufflation of CO₂ through the supraumbilical incision with visiport insertion of 11 mm, 11 mm in the right lumbar region and another 5 mm in left lumbar region was done, the peritoneal flap was dissected with the creation of upper and lower flaps, hernia sac was then isolated, and vas was identified. During sac dissection, a hard mass arising from the left cord (that was thought of as a sac) was discovered. Dissection was performed to release the mass from the cord structures and isolate the vas. Surgery was carried on with mesh insertion and closure of the flaps with suture v lock. The rest of the abdomen was inspected and looked clear at the time of surgery.

Postoperative recovery went smoothly, and the patient was discharged on the next day with outpatient follow-up. Postoperative pathological examination revealed a picture of...
malignant growth of atypical cells lining the tunica vaginalis with multiple mitotic (Figs. 2a-2d), and multinucleated atypical nuclei were seen, diffuse immunoreactivity for mesothelial markers, including calretinin, cytokeratin 5/6, and Wilms’ tumor gene 1, was evident (Fig. 3) and a diagnosis of malignant mesothelioma arising from the left tunica vaginalis testis was made.

Fig. (1). The left groin shows an ill-defined mixed echogenicity, predominantly hypoechoic linear lesion surrounded by echogenic material, extending into the left scrotal sac, consistent with inguinal scrotal hernia.
Histological sections showed characteristic features of malignant mesothelioma of the tunica vaginalis. These sections illustrated malignant growth forming multinodular proliferation infiltrating the soft tissue and lining the thickened tunica and infiltrating with papillary and tubulopapillary pattern. Lined by single to stratified atypical mesothelial cells with abundant pink epithelioid cytoplasm. Enlarged single to multinucleated atypical, hyperchromatic nuclei with prominent nucleoli, occasional mitotic figures. Many tumor giant cells, few psammoma bodies seen. There is no spindled or biphasic pattern. The malignant cells seen infiltrating in solid nests into the fibrofatty tissue with dense chronic inflammation, foamy cells, brown pigmentation and lymphoid follicles. Focal areas of necrosis also seen with desmoplasia.

Immunohistochemical stains of malignant mesothelioma of tunica vaginalis illustrated expression of CK 5/6, Calretinin, Ki 67 (proliferative index 6-8%), and its negative for CEA, PR.
The patient was then referred to the oncology center, where cytoreductive surgery with hyperthermic intraperitoneal chemotheraphy (CRS-HIPEC) was performed and 4 cycles of chemotherapy were given including (Cisplatin, Pemetrexed and Bevacizumab) in April 2019 with last follow CT scan done on September 2019, which was negative.

4. DISCUSSION

Testicular cancer accounts for around 1% of all male tumors, where the incidence rate has been increasing and doubling over the last decade. Around half of the cases occurred between the ages of 20 and 34 and approximately 85% of testicular tumors were germ cell tumors and the rest were paratesticular and gonadal cell tumors. Comparing all testicular tumors, 40.7% were seminomas while 44.6% were nonseminomas, however, the incidence rate for the subtypes of the nonseminomas was: a mixed tumor in 51.6%, embryonal carcinoma in 19.9%, yolk sac tumor in 12.3%, germinomas in 6.7%, teratomas in 6%, and choriocarcinomas in 3.6% where the incidence of paratesticular tumor group was: lymphomas (34.7%) and rhabdomyosarcomas (23.6%) [3].

The 5-year survival rate is more than 95% of the germ cell tumors as it is one of the most curable forms when diagnosed earlier [4]. Around 76% of stage I seminoma relapses occurred within the first two years, where, 70% of stage I nonseminoma relapses occurred within six months [5].

The first case of malignant testicular mesothelioma in the literature was reported by Barbera and Rubino in 1957 [6], and as mentioned by Zhang and Fu, around 100 cases have been reported up to that date [7].

The etiology and development of malignant mesothelioma have not been fully understood [7], it is known that it is caused by a mutation of mesothelial cells that make up the lining of organs including lung pleura, pericardium, peritoneum and tunica vaginalis that lines the testes. The disease makes the testicles lining thicker, producing the fluid buildup. It can affect males of any age but most commonly those aged 55 – 75 years [8]. Exposure to asbestos can be of any duration and maybe for several years in the past is a known risk factor that significantly increases the risk of developing malignant mesothelioma [9]; long term hydrocele, inguinal hernia and paratesticular mass have also been frequently associated with testicular mesothelioma [10]. Our case included a patient who had a history of long-term hydrocele. Preoperative diagnosis is very challenging and unusual; however, this tumor is very aggressive, testicular swelling is the most common but unspecific symptom. Diagnosis should be made in any patient with a history of exposure to asbestos presenting with the testicular disease [11].

Clinical evaluation including physical examination and imaging plays an important role in suspecting and diagnosing testicular tumors, in addition, pathological evaluation is very essential in diagnosing the different types of testicular tumors even the rare ones like testicular mesothelioma. According to histopathology, malignant mesothelioma has been sub-classified as epithelioid, sarcomatoid and biphasic, and epithelial type is the one most commonly associated with malignant testicular mesothelioma [8]. It is also graded as a high- and low-grade tumor according to stromal and vascular invasion, atypia and lymphocyte infiltration. Immunohistochemistry also greatly aids in the accurate diagnosis. Patient in our case was younger than the average age, had a low-grade tumor, and diffuse immunoreactivity for mesothelial markers, including calretinin, cytokeratin 5/6, and Wilms’ tumor gene 1, was evident but staining for placental alkaline phosphatase (a marker of seminomas) and alpha-inhibin (a marker of sex cord stromal tumor) was negative.

Treatment of malignant testicular mesothelioma involves surgery, chemotherapy and radiotherapy; combination therapy can be used according to the tumor stage and the presence of metastasis. In our case, the left orchiectomy was performed combined with HIPEC therapy.

CONCLUSION

Testicular malignant mesothelioma is an extremely rare tumor, it is an invasive and often fatal disease, the clinical presentation is unspecific, and diagnosis is very difficult and challenging. When a testicular swelling or mass is encountered, a detailed history and physical examinations are required, a variety of causes should be considered, and the diagnosis of the testicular tumor could suspect even rare tumors like testicular mesothelioma.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This study is approved by the Ministry of Health and Prevention Research Ethics Committee under reference number MOHAP/DXB-REC/FFF/NO.21/2020.

HUMAN AND ANIMAL RIGHTS

Not applicable.

CONSENT FOR PUBLICATION

Written and informed consent has been obtained from the patient for this study.

STANDARD FOR REPORTING

CARE guidelines have been followed in this case report.

FUNDING

None.

CONFLICT OF INTEREST

The authors declare no conflict of interest, financial or otherwise.

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