

Hydrocele with a surprise: Malignant mesothelioma of the tunica vaginalis - Case report and review of literature

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Abstract

Hydrocele is a very common condition that is simple to evaluate and treat. Management of hydrocele is usually delegated to the junior members of the surgical team. Sometimes this simple condition can spring huge surprises. A 20-year-old man presented with acute onset large, painless fluctuant left hemi-scrotal swelling. Scrotal ultrasonography showed thickened tunica vaginalis. A diagnosis of left hydrocele was made and repair by excision of sac was planned. During the procedure, the sac was found studded with red nodular growths; histopathology reported malignant mesothelioma of tunica vaginalis. Metastatic evaluation showed extensive retroperitoneal lymph nodal involvement. Despite receiving adjuvant chemotherapy with radiotherapy patient died due to extensive metastasis within 16 months. This case is presented for rarity of diagnosis, young age of presentation, absence of etiological factor and rapidly progressive clinical course.

Key Words: Chemotherapy, radical orchiectomy, radiotherapy

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INTRODUCTION

Hydroceles are one of the common causes of scrotal swellings often with a benign course.^[1] Thus, with the dictum, "Common things occur commonly" hydrocele may be presumed to be benign until proven otherwise. Usually, it is a simple condition to manage, but sometimes, there can be huge surprises. Such huge surprise can be an incidental finding of malignant hydrocele in form of mesothelioma of the tunica vaginalis, a rare lethal and aggressive disease which in most cases are reported at 55–75 years of age with only 10% of cases seen

in those younger than 25 years.^[2] It was first described by Barbera and Rubino in 1957,^[3] and by 2010, about 223 cases have so far been reported.^[4] Exposure to asbestos is a known risk factor.^[2,4,5] Most common presentation is hydrocele, and diagnosis is almost always intraoperative.^[2] Radical orchiectomy with adjuvant radiotherapy and chemotherapy is treatment of choice, but overall prognosis is very poor with median survival of 23 months.^[2]

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MATERIALS AND METHODS

Twenty-year-old Indian male patient presented with a 3 weeks history of painless swelling of the left hemiscrotum. There was no fever, trauma, or lower urinary tract symptoms. Examination revealed a fluctuant, nontender, and 10 cm left hemiscrotal swelling in its longitudinal axis with normal scrotal skin. Baseline investigations were normal. Ultrasound showed normal testis with irregular thickening of sac wall. Excision of the hydrocele sac was planned through conventional scrotal approach.

Intraoperative findings showed a thick vascular hydrocele sac filled with amber-colored fluid. The parietal surface of the tunica vaginalis was lined with numerous red firm vascular nodules of 15–18 mm [Figure 1]. Testis and the epididymis were normal.

Histopathology of the tunica vaginalis revealed malignant mesothelioma of the tunica vaginalis.

On further evaluation of the patient, there was to exposure to asbestos and computerized tomography of the abdominopelvic region showed large left para-aortic and left renal hilar lymph nodes (4 cm × 3 cm). The patient underwent radical orchiectomy with excision of scrotal scar. Ten days postoperation, there was a large recurrence of growth (6 cm × 4 cm) and extensive involvement of the scrotal skin [Figure 2].

Adriamycin-based chemotherapy with local radiotherapy was started. There was an initial partial response, but the patient steadily deteriorated and died within 16 months with extensive metastasis.

DISCUSSION

Malignant mesothelioma of the tunica vaginalis is a very rare neoplasm associated with highly aggressive biological

behavior.^[6] It commonly occurs in elderly age group with a rapidly progressive course. Few cases have been reported in the younger age group of <25 years^[2] as it was observed in the index case reported.

Exposure to asbestos is considered a risk factor for its development^[2,4,5] with some stray reports of malignant mesothelioma developing in long duration hydrocele.^[7] Furthermore, trauma and herniorrhaphy have been reported as other possible predisposing factors.^[8] However, not all reported cases of mesothelioma of the tunica vaginalis were associated with exposure to asbestos.^[8,9]

Most patients with localized disease present with nondisease specific features such as hydroceles, epididymitis, spermatoceles, and scrotal hernia which make preoperative diagnosis difficult.^[2,5] Advanced disease may be associated with para-aortic lymph nodal involvement and inguinal mass. Metastasis to the viscera may occur (liver and lungs).^[5]

Nonspecific presentations and rarity of the disease make preoperative diagnosis difficult. Majority of reported cases (97.3%) were diagnosed intraoperatively or postoperatively.^[2,10]

Preoperative diagnosis could be suspected in a much older patient (55–75 years) with history of exposure to asbestos or long duration of hydroceles and with scrotal ultrasonography finding of focal nodularities on hydrocele sac.^[11]

Ultrasound-guided fine-needle aspiration cytology (FNAC) could confirm the diagnosis. Although cytology of hydrocele fluid may not give adequate yield, FNAC of the focal nodularities instead of the fluid from the hydrocele may improve the yield.^[12] Furthermore, scrotal magnetic resonance imaging may demonstrate nodular masses with irregular surfaces lining of the hydrocele cavity.^[11]



Figure 1: Computed tomography imaging showing thickened parietal surface of the tunica vaginalis



Figure 2: Radical orchiectomy specimen

In the index case report, the patient was of much younger age group with no history of exposure to asbestos and short duration of hydroceles; scrotal ultrasound only revealed a thickened tunica vaginalis. These atypical presentations did not suggest mesothelioma of the tunica vaginalis; diagnosis was made by histology postexcision of the hydrocele sac riddled with numerous vascular nodules.

The management of malignant mesothelioma of the tunica vaginalis requires staging of the disease to determine its extent with subsequent aggressive surgery and adjuvant therapy to achieve long-term survival.^[2,13]

Radical orchidectomy for local disease control and where there is violation of the scrotal wall, wide local resection of scrotum, or hemiscrotectomy is required.^[2] This is usually followed up by adjuvant radiotherapy and/or chemotherapy. Radiotherapy was reported to be more effective than chemotherapy.^[2] Albeit, adjuvant therapy with antineoplastic chemotherapy or radiation therapy may be disappointing with their benefits reported to be negligible.^[13,14]

For localized disease, the following protocols have been suggested:^[13]

- Initial staging of suspected cases with computed tomography of the abdomen and pelvis
- Radical inguinal orchiectomy or hemiscrotectomy
- Retroperitoneal lymph node dissection in cases with positive nodes on scan or biopsy
- Inguinal node dissection in cases requiring hemiscrotectomy.

For advanced or recurrent disease, local radical resection with chemotherapy, including high-dose cisplatin and doxorubicin for two cycles of 5 days each with local radiotherapy for uncontrolled locally advanced disease, is advised.^[13]

Patients with localized disease have been known to survive for more than 10 years.^[6] However, majority will not live beyond 5 years, with median survival being 23 months.^[2]

Follow-up of the patient after initial treatment is important as malignant mesotheliomas of the tunica vaginalis are prone to local recurrence, lymphatic, and hematogenous dissemination. Overall recurrence rate (local and disseminated) was 52.5%. More than 60% of the recurrences developed within first 2 years of the follow-up.^[2]

CONCLUSION

This case was reported due to rarity of mesothelioma of the tunica vaginalis, diagnosed in a young patient with clinical presentation of hydrocele, and no associated risk factors; subsequently, manifested rapidly progressive clinical course with lethal, aggressive behavior. Awareness of the disease with an aim for accurate preoperative diagnosis may improve the outcome of this disease.

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Conflicts of interest

There are no conflicts of interest.

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