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Subcutaneous leiomyosarcoma of the scrotum mimickering testicular cancer. A case report and literature review

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ABSTRACT

Leiomyosarcoma of the scrotum is a rare tumor. Johnson H Jr in 1987 reported first case. Only 39 cases been reported in the literature. A 74 years old Indian gentleman presented with progressive painless right testicular swelling for 3 months. On examination, a firm right testicular mass. We proceeded with right scrotal exploration with excision of the right scrotal mass. Histopathology showed scrotum leiomyosarcoma. We would like to discuss the rarity of presentation, dilemma of diagnosis and managing in a district hospital. We advocate wide local excision with clear margin of 2 cm which give good overall survival benefits.

Introduction

Leiomyosarcoma of the scrotum is a rare type of tumor especially with no involvement of the testis, epididymis, or spermatic cord. This known as subcutaneous superficial leiomyosarcomas. Diagnosing leiomyosarcomas of the scrotum is such that a General Practitioner only sees once in every 20 years. In view of only 39 cases are been reported, most practitioners would never come across this tumor in their lives.

Case presentation

A 74 years Indian gentleman presented with history of progressive painless right testicular swelling for 3 months. Upon examination, there was a multilobulated, painless firm mass at the right testicular region measuring 5 × 6cm with erythematous skin changes with ulceration. Ultrasound scrotum showed a large heterogeneous hypoechoic solid mass measuring 6.4 cm × 8.0 cm (AP x Wt) within the right scrotum as shown in Fig. 1. The tumor markers β-HCG, α-fetoprotein was normal. We proceeded with right scrotal exploration with excision of the right scrotal mass. Intraoperative showed right scrotal mass measuring 8 × 6cm, hard, multilobulated and well vascularised and was able to separate from the right testis and spermatic cord as shown as Fig. 2. Histopathology showed fairly circumscribed cellular tumor with area of haemorhage and necrosis. Tumor composed of interlacing bundles of spindle to plump spindle nuclei with whorling in areas. The tumor cells are pleomorphic, having round to oval, vesicular to hypochromatic nuclei, coarse chromatin with inconspicuous nucleoli suggestive of leiomyosarcoma of right scrotum. Immunohistochemically, vimentin, smooth muscle actin (SMA) and desmin was positive meanwhile S-100 was negative. Post operatively we proceed with CT TAP there was no distant metastasis.

Discussion

Soft tissue Sarcomas constitute 1% of all malignancies. Leiomyosarcomas comprise of 10% of soft tissue sarcomas. Leiomyosarcomas arise common from uterus, gastrointestinal tract and retroperitoneal region. Subcutaneous leiomyosarcomas (LMS) comprise of 1% of all superficial soft tissue malignancies. Leiomyosarcoma of scrotum are malignant mesenchymal neoplasm's arising from smooth muscle linings of arterioles and veins in the subcutaneous layer. To date about 39 cases of leiomyosarcoma of scrotum have been reported in literature.[1] Johnson H Jr first reported LMS of scrotum in 1987. Scrotal LMS present between the 4th and 8th decades of life as a painless, slow-growing skin lesion of size 2–9 cm.[2] However, our patient presented with progressive painless scrotal mass within 3 months duration. Leiomysarcomas of the scrotum present as firm, rubbery, non tender, irregular masses which can be confused as testicular tumor. Laboratory investigation are important to exclude germ cell tumor, example if β-HCG and α-fetoprotein are normal The role of biopsy is important for confirmation of diagnosis of leiomyosarcoma. Histological, there composed of interlacing bundles of spindle to plump spindle nuclei with whorling in areas as shown in Fig. 3. Immunohistochemistry is important to differential between leiomyosarcomas and other rare tumors, such as benign

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leiomyoma, fibrous mesothelioma, and various benign fibrous tumors. Leiomyosarcomas are positive for actin, desmin and CD 34. Japanese guidelines for the treatment of soft tissue tumor, recommend that soft tissue tumors should be resected together with a 2 cm margin. Wide excision of scrotal leiomyosarcomas with a clear margin of at least 10 mm was associated with a better outcome in comparison to tumors with involved margin or less than 10 mm clear margin3. Currently there is no clear indication for prophylactic lymphadenectomy. There are no available studies which demonstrating that prophylactic lymph node dissection provides significant survival or recurrence benefit for patients.

The tumor cells are pleomorphic, having round to oval, vesicular to hypochromatic nuclei, coarse chromatin with inconspicuous nucleoli. The role of adjuvant treatment is not considered in all cases and its controversial. Chemotherapy may be commerce in patients who refuse surgery. Chemotherapy regime are gemcitabine, paclitaxel, vincristine, doxorubicin, actinomycin-D has shown limited success in treating leiomyosarcoma. Radiotherapy has been of doubtful value in Leiomyosarcoma except for palliative patients.[4] The mode of spread of leiomyosarcoma is primarily haematogenous to lung, liver and bone. The prognosis of leiomyosarcoma depends upon the size, depth and grade of the tumor and evidence of distant metastases. In our case our patient did not have any distant metastasis. Recurrence most commonly tends to be local but distant metastases in the bones and lungs have been reported. Nonetheless, long term follow up of patients is mandatory to detect local recurrence and distant metastases that can occur years after the initial excision. In view of only about 39 cases are reported it would be argued that most practitioners would never come across a scrotal leiomyosarcoma in their lives. However, they should refer such cases to tertiary hospital with urology specialty.

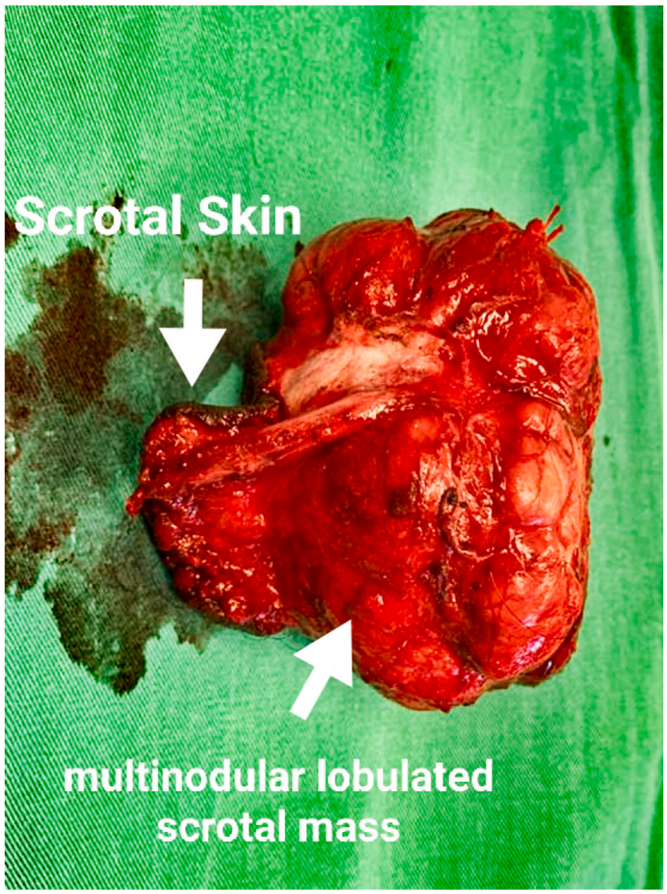

Fig. 2. Right scrotal mass measuring 8 × 6cm,hard, multilobulated and well vascularised. Clear demarcation between the right testis together with its cord with the scrotal mass. Right testis normal in size and shape. No hydrocele.

Conclusion

Leiomyosarcoma of scrotum should be considered as a differential diagnosis in any elderly male presenting with an scrotal mass. We advocate that surgical management still the mainstay. A wide local excision with clear margin of 2 cm. Adjuvant chemoradiotherapy is still controversial and doesn't give any overall survival benefits. The limited number of cases of this rare disease as well as the inconsistent

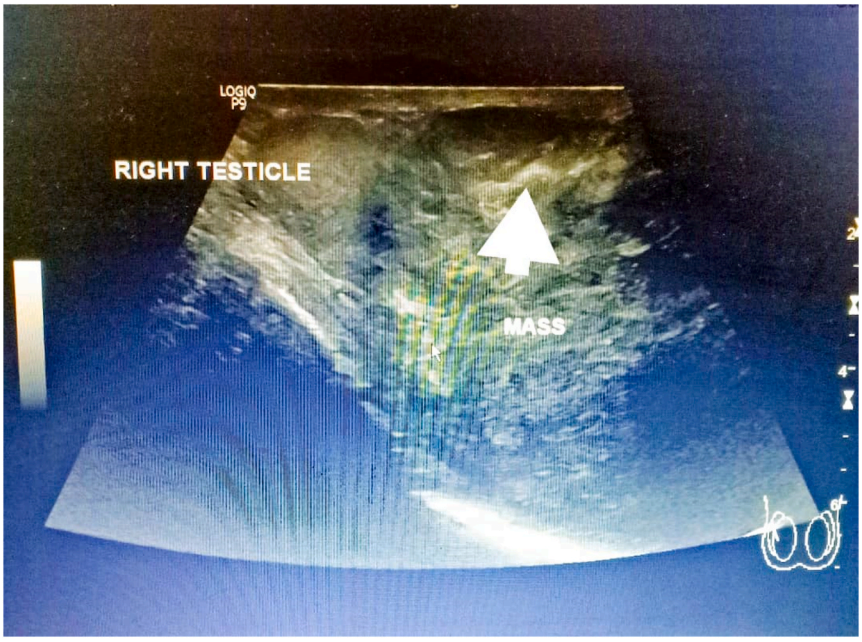

Fig. 1. A large heterogenous hypoechoic solid mass is seen within the right scrotum, inferior to the right testis. It measuring 6.4 cm × 8.0 cm (AP x Wt). Clear plane of demarcation is seen between this mass and the adjacent right testis.

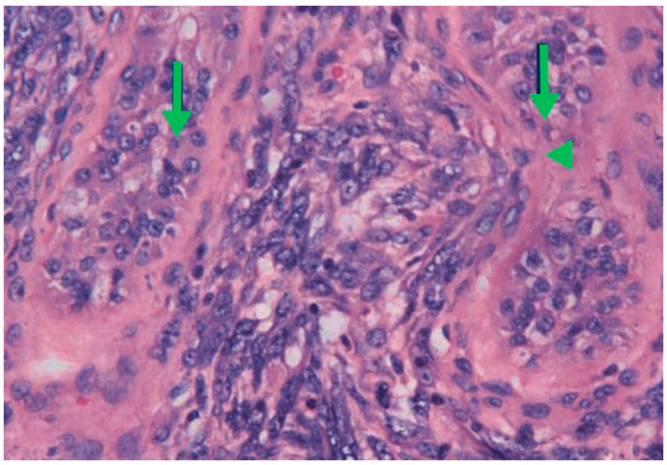

Fig. 3. Tumor composed of interlacing bundles of spindle to plump spindle nuclei with whorling in areas.

management requires that further research be performed to formulate an ideal treatment protocol and strategies.

Declaration of competing interest

We declare that this manuscript is original, has not been published before and is not currently being considered for publication elsewhere.

We know of **no conflicts of interest** associated with this publication, and there has been no significant financial support for this work that could have influenced its outcome.

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