

Recurrent Leiomyosarcoma is a Rare Commodity in Penile Cancer: A Review and Report

P Jayakumar^{1*}, Manish² and MD Ray³

¹Senior Resident, Department of Surgical Oncology, Dr. BRA Institute Rotary Cancer Hospital, All India Institute of Medical Sciences, New Delhi

²Assistant professor, IRCH, AIIMS, New Delhi, India

³Associate professor, IRCH, AIIMS, New Delhi, India

***Corresponding author:** Jayakumar P, Department of Surgical Oncology, Dr. BRA Institute Rotary Cancer Hospital, All India Institute of Medical Sciences, New Delhi

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Abstract

Squamous cell carcinoma constitutes approximately 95% of penile cancer. Sarcomas of penis are rarer among them. Leiomyosarcoma is one of the extremely rare sarcoma of penis. To best of our knowledge and literature review, only few cases have been reported world-wide so far. We are presenting this uncommon case of recurrent leiomyosarcoma of penis in a 37-year male patient which was diagnosed with microscopy with its characteristic immunohistochemistry along with a concise review.

Keywords

Squamous cell; Leiomyosarcoma; leiomyosarcoma.

Introduction

Case History

A 37-year male patient presented to the Surgical Oncology OPD with complaint of a recurrent nodular growth at the glans penis, which was operated multiples times in various time period. The patient was

noted the swelling on 2015 and underwent WLE in a government medical college and hospital on 2016. The final HPR reported as malignant mesenchymal tumour of penis, IHC showed positive for Vimentin, SMA & Desmin suggestive of leiomyosarcoma of penis Grade 2, the resected margin turned to be positive. So, patient underwent Re-WLE with 1 cm margin. The disease-free interval was 5 years. Again, patient developed nodular swelling over the ventral aspect of the glans penis for one year. Patient underwent incisional biopsy showed features of cutaneous leiomyosarcoma. There was no inguinal lymphadenopathy. Routine laboratory work up showed a normal picture. Patient underwent CECT chest, abdomen and pelvis showed only few sub centimetric inguinal lymphnodes, no lung and distant metastasis. The intraoperative findings was 1x1 cm nodular swelling present over ventral aspect at the coronal sulcus at 6'o clock reaching till corpora spongiosum, urethra and overlying skin free of tumour. The patient underwent a partial glansectomy along with prepuce and neo-ureterostomy with 0.5cm margin all around tumor. The part of glans and external urethral meatus were excised for margin. The histopathology report shows a nodule measuring 0.6x0.4cm cut section is grey-white involving the dermis of glans penis. Multiple sections examined show features of a cutaneous Leiomyosarcoma. All margins are free of tumor.



Figure 1: Neo-ureterostomy over corona of glans on ventral aspect like hypospadiasis.



Figure 2: Catheter placed through neoureterostomy.



Figure 3: Resected specimen with tumour.

Review and Discussion

The squamous cell carcinoma is the most common primary malignant neoplasm of the penis. Rarely, metastatic neoplasms extending from prostate, bladder, rectum, kidney and testis can be occurring. Sarcomas are rare and they constitute less than 5% of all types of penile malignancies. Few sarcomas like Kaposi sarcoma, epithelioid hemangioendothelioma and angiosarcoma, followed by rhabdomyosarcoma and leiomyosarcoma are common in penis [1].

There are a number of potential sources within the penis from which leiomyosarcoma can arise: 1) the dartos muscle layer of the prepuce and shaft, 2) the arrector pili muscles associated with lanugo hairs on the penile shaft, 3) the muscular walls of superficial vessels situated outside of the tunic albuginea, and 4) the muscular walls of the deep vascular complex that make up the corpus cavernosum and corpus spongiosum. There are two distinct clinico-pathological entities of leiomyosarcoma, superficial and deep-seated tumors. The cutaneous leiomyosarcoma present as small nodular growth, appears more distal, slow growing and have a low metastatic potential. They arise from the muscle fibres of the dartos, erect or pili and superficial vessels in tunica albuginea. Superficial LMS grow slowly and have more tendency to recur locally although distant metastasis is uncommon. On the other hand, Deep-seated lesions originate from the corpus spongiosum and these tend to metastasize early or they invade the urethra. The differential diagnosis for leiomyosarcoma of the penis includes leiomyoma, myointimoma, nodular Kaposi sarcoma, malignant fibrous histiocytoma, and sarcomatoid carcinoma. Leiomyomas of the penis are very rare, perhaps even rarer than leiomyosarcomas. These tumors tend to present as small, painless, slow-growing masses in mid adult life. Histologically, they are typically well demarcated, and in contrast with leiomyosarcomas, they should not have cytologic atypia and mitotic activity [2].

The superficial type of leiomyosarcoma has a good prognosis with only wide local excision, partial penectomy or total penectomy. Distant metastasis is rarely observed. The deep type of leiomyosarcoma requires more aggressive treatments, including radical penectomy, radiation therapy, and chemotherapy, although the outcome of these treatments is still poor. In one study, it was reported that 69% (9/13) of patients died within 3 years after diagnosis because of multiple metastases and dissemination of the disease. Excision of the regional lymph nodes in deep type leiomyosarcoma has no definite benefits for a patient's survival contrary to squamous cell carcinoma of the penis. Also, radiation therapy and chemotherapy produce little effect in treating leiomyosarcoma of the penis, but they might decrease the risk of local and distant recurrence postoperatively. Accepted poor prognostic factors of leiomyosarcoma include deep tumor depth, a tumor size of greater than 5 cm, a high mitotic rate, and necrosis [3].

On gross examination, these tumours are usually rubbery in consistency, well circumscribed, with a white, yellow or grey appearance sometimes shows capsulation. It has been stated that gross examination in cases of leiomyosarcoma does show tumor that has measured between 0.5 cm and 6.0 cm and that the median size has been 1.5 cm. It has been documented that leiomyosarcoma have been reported most often been superficial tumors and does reveal tumors that are white-tan-grey, and firm as well as the tumors tends to have irregular borders [4].

The microscopic examination features of the tumor do show interlacing fascicles of spindled-cells that have abundant eosinophilic cytoplasm as well as focal juxta-nuclear vacuoles and blunt ended nuclei. Microscopic examination of leiomyosarcoma also does show a degree of atypia which tends to range between moderate atypia to severe atypia. Microscopic examination of leiomyosarcoma on rare occasions does show atypical multi-nucleated giant cells, and focal necrosis.

- **Immunohistochemistry studies positive staining:** The IHC staining studies in cases of leiomyosarcoma tend to show that the tumors do exhibit positive staining for the ensuing tumor markers: Desmin (positive staining), Muscle specific actin (positive staining). Alpha smooth muscle actin (positive staining). The tumor cells would also tend to exhibit positive staining for vimentin.
- **Electron microscopic examination:** It has been stated that electron microscopy examination of leiomyosarcoma of the penis will shows Myofibrils, dense bodies, and abundant pinocytic vesicles tend to be noted around majority of the tumor cells.
- **Kaposi sarcoma (Nodular):** These tumors tend to be located superficially and microscopic examination of the Kaposi Sarcoma tumors tend to show slit-like spaces associated with many erythrocytes, no evidence of atypia. Immunohistochemistry staining studies of Kaposi sarcoma of the penis tends to show positive staining for: HHV8, CD31, CD34, Factor VIII.
- **Leiomyoma:** Leiomyomas of the penis are extremely rare and the microscopic examination features of these benign lesions tend to be similar to the features of leiomyoma's elsewhere within the body.
- **Melanoma with storiform growth:** Melanoma with storiform growth within the penis does look like typical melanoma elsewhere within the body. Immunohistochemistry staining studies of melanoma with storiform growth pattern does show positive staining for melanocyte markers including: HMB45, Melan A.
- **Sarcomatoid carcinoma:** Sarcomatoid carcinoma of the penis does consist of squamous epithelial nests within the tumor mass and associated areas of PeIN. Immunohistochemistry staining studies in sarcomatoid carcinoma of the penis does exhibit positive staining for: Pankeratin, Keratin 34beta E12 and p63. In sarcomatoid carcinoma of the penis immunohistochemistry staining studies show negative staining for muscle markers [5].

Conclusion

Primary noninvasive leiomyosarcomas of the penis is a rare commodity of penile cancer. They would tend to have good outcome following complete surgical excision but in case of large noninvasive tumors with high mitotic activity, could subsequently recur regular follow up is advisable.

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