Case Report

Primary Penile Leiomyosarcoma of Penis: A Rare Case Report

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Abstract

Penile tumors rarely include mesenchymal tumors, and within this group, sarcomas are even more uncommon. We report a case of primary leiomyosarcoma of the penis in a 45-year-old male who underwent total penectomy for an ulcero-proliferative mass involving the distal shaft and extending up to the base of the penis. Grossly, the tumor measured 10 × 5 × 5 cm. Microscopic examination revealed spindle cells arranged in fascicles with cigar-shaped nuclei and marked atypia. Immunohistochemistry showed positivity for vimentin, smooth muscle actin, and h-caldesmon, confirming smooth muscle origin. The histopathological examination and immunohistochemical workup remain the gold standard for rendering this diagnosis at such a rare site. This case emphasizes the importance of accurate diagnosis and complete surgical excision, along with long-term follow-up for optimal patient outcomes.

Keywords: Histopathology; Spindle cell sarcoma; Immunohistochemistry; Mesenchymal tumour

Introduction

Primary leiomyosarcoma of the penis is an uncommon malignant neoplasm arising from smooth muscle elements, accounting for less than 0.1% of all penile malignancies [1, 2]. While squamous cell carcinoma is the predominant penile cancer, sarcomas such as leiomyosarcoma, although rare, should be considered in the differential diagnosis of non-epithelial penile masses [2, 3]. Approximately 60-61 cases have been documented in the English literature to date, with the most recent comprehensive review in 2023 reporting 61 cases [4, 5]. These tumors may present as either superficial or deep lesions, with the latter often exhibiting a more aggressive clinical course [6]. Given its rarity, diagnosis and management are often delayed or misdirected, highlighting the need for increased clinical awareness [7].

Case Report

A 45-year-old male presented with an ulcero-proliferative lesion over the penis for the past 5 months, which was increasing in size and accompanied with pain. Clinical examination revealed a mass involving the distal shaft, glans, and urethra, extending proximally toward the penile base. The lesion was clinically suspected to be squamous cell carcinoma. Bilateral inguino-pelvic lymphadenopathy was noted on palpation.

Routine biochemical and haematological investigations were within reference ranges. Chest X-ray and abdominal ultrasonography showed no abnormalities. A contrast-enhanced computed tomography (CECT) scan of the chest revealed no evidence

of distant metastasis. A wedge biopsy performed at an outside facility initially suggested a diagnosis of well-differentiated squamous cell carcinoma.

The patient subsequently underwent a total penectomy, along with bilateral inguino-pelvic lymph node dissection and perineal urethrostomy. Gross pathological examination revealed an ulcero-proliferative lesion measuring 10 × 5 × 5 cm involving the distal penile shaft and glans, extending close to the penile base (Fig 1). The tumor was well-circumscribed and involved the corpus spongiosum and glans. The overlying skin showed ulceration. Multiple sections were taken from the tumor, surrounding penile tissue, urethral margin, and proximal surgical margin. All surgical margins were inked before sectioning. The cut surface of the tumor appeared firm and grey-white.

On microscopic examination of the hematoxylin and eosin stained sections, the tumor cells were arranged in long and short fascicles of spindle cells with eosinophilic cytoplasm and hyperchromatic elongated cigar-shaped nuclei with blunt ends exhibiting marked atypia. Mitoses were 0-2 per 10 high power fields (HPF diameter: 0.55 mm). Focal areas of tumor necrosis were noted. No lymphovascular or perineural invasion was identified. The tumor was located 3 mm from the urethral mucosa but did not invade it. All examined surgical margins were free of tumor. The tumor was graded as Grade 1 (FNCLCC grading system: Differentiation score 2, Mitotic count score 1, Necrosis score 1; Total score 4/8). Bilateral inguinal lymph nodes (12 nodes examined) showed reactive hyperplasia with no evidence of metastasis.

Based on these histopathological features, differential diagnoses considered included leiomyosarcoma, sarcomatoid carcinoma, spindle cell melanoma, and other spindle cell sarcomas. To confirm the diagnosis and rule out these differentials, immunohistochemistry was performed.

Immunohistochemistry confirmed the diagnosis: tumor cells were diffusely positive for vimentin, smooth muscle actin (SMA), and h-caldesmon, while negative for cytokeratin and desmin (Fig 2 and 3). Additionally, CD34, S100, HMB-45, and CD68 were negative, thereby excluding other sarcomas, melanoma, and histiocytic lesions. The immunohistochemical panel details are as follows: Vimentin (clone V9, Dako, 1:100), SMA (clone 1A4, Dako, 1:200), h-Caldesmon (clone h-CD, Dako, 1:100), Desmin (clone D33, Dako, 1:100), Pan-cytokeratin (AE1/AE3, Dako, 1:100), CD34 (clone QBEnd10, Dako, 1:50), S100 (polyclonal, Dako, 1:400), HMB-45 (clone HMB45, Dako, 1:50), and CD68 (clone KP1, Dako, 1:100). Detection was performed using the EnVision FLEX detection system (Dako). Appropriate positive and negative controls were run with each batch. Table 1 summarizes the immunohistochemical findings that helped differentiate leiomyosarcoma from its mimics.

The postoperative course was uneventful. Unfortunately, the patient left the hospital against medical advice, and further oncological management and follow-up were not possible. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

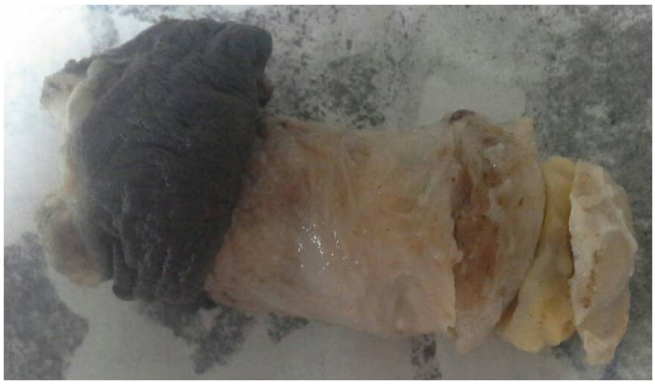

Figure 1: Gross photograph of total penectomy specimen showing ulceroproliferative growth measuring 10 × 5 × 5 cm.

Discussion

Primary leiomyosarcoma (LMS) of the penis is an exceedingly rare mesenchymal neoplasm, with approximately 60-61 well-documented cases reported worldwide as of 2023 [4, 5]. It originates from the smooth muscle elements of penile tissue, including the dartos muscle, vessel walls, and erectile tissue [5]. Clinically, these tumors often mimic other spindle cell lesions, and misdiagnosis is common—particularly when presenting as ulceroproliferative growths, which are often presumed to be squamous cell carcinoma, as in our case [7].

Microscopic examination of LMS reveals spindle cells arranged in intersecting fascicles, which have eosinophilic cytoplasm and blunt-ended, ”cigar-shaped” nuclei. A low mitotic index may be misleading and must be correlated with atypia and necrosis for accurate grading. Immunohistochemistry (IHC) is a key tool for differentiating LMS from its morphologic mimics [4]. Our case showed positivity for SMA and h-caldesmon, confirming smooth muscle lineage, and was negative for

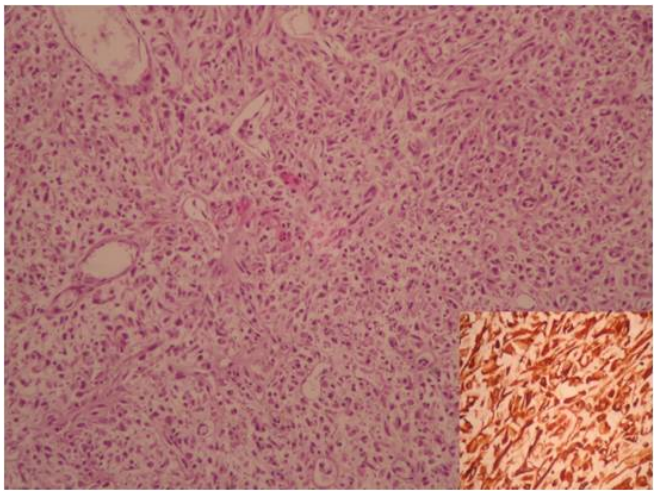

Figure 2: Spindle cells in fascicles with eosinophilic cytoplasm and cigar-shaped nuclei (H&E, 400×). Inset: diffuse vimentin positivity (DAB, 400×).

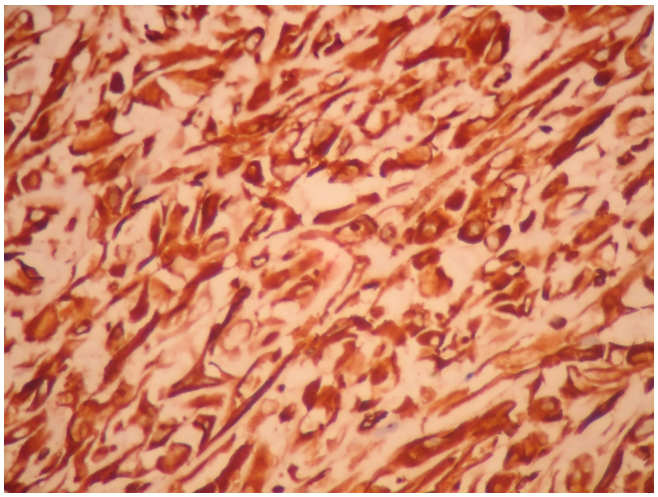

Figure 3: Diffuse smooth muscle actin immunopositivity (DAB, 400×).

cytokeratin and desmin, helping to exclude sarcomatoid carcinoma and rhabdomyosarcoma, respectively.

Spindle cell lesions of the penis must be distinguished from entities such as sarcomatoid carcinoma, Kaposi sarcoma, and melanoma. These entities can mimic LMS histologically but have distinct IHC signatures and morphologic features as described in Table 1.

Table 1: Histomorphological and immunohistochemical features of spindle cell tumors.

Entity	IHC Profile	Morphologic Clues
Leiomyosarcoma	SMA+, h-caldesmon+, Desmin variable, Vimentin+	Cigar-shaped nuclei, fascicular growth
Sarcomatoid carcinoma	Keratin+	Epithelial component, severe atypia
Kaposi Sarcoma	CD34+, HHV8+	Slit-like vessels, extravasated RBCs
Melanoma	S100+, HMB45+	Prominent nucleoli, presence of melanin pigment

The accurate diagnosis of LMS relies on an integrated histopathologic and immunophenotypic approach, especially in uncommon sites such as the penis [4]. Deep LMS variants are often associated with a higher propensity for recurrence and metastasis [6], making early and accurate diagnosis crucial for surgical planning and prognosis. Reported recurrence rates for penile leiomyosarcoma range from 30-50% in published series, with local recurrence being more common than distant metastasis [4, 6]. This underscores the critical importance of achieving negative surgical margins and establishing long-term follow-up protocols. In our case, the patient's loss to follow-up represents a significant limitation in assessing treatment outcomes and detecting potential recurrence.

The primary treatment for penile LMS involves surgical excision to achieve negative margins [4]. The role of adjuvant therapy is not well defined due to the paucity of cases, although radiotherapy and chemotherapy may be considered in

high-grade or metastatic tumor [6]. Unfortunately, our patient was lost to follow-up before any further oncological evaluation could be undertaken.

Conclusion

Primary penile leiomyosarcoma is an extremely rare entity that can mimic more common penile malignancies both clinically and histologically. A high index of suspicion, along with the use of appropriate immunohistochemical markers, is essential for correct diagnosis. Early recognition and complete surgical excision are key to improving patient outcomes. This case underscores the importance of multidisciplinary coordination in managing rare penile tumors.

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