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Case Report

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STUMPED: pelvic smooth muscle tumour of uncertain malignant potential in a male

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ABSTRACT

Smooth muscle tumours of uncertain malignant potential (STUMPs) are rare neoplasms that present diagnostic difficulties due to their intermediate morphology. While they represent an uncommon uterine tumour type, extra-uterine especially in males, are exceptionally rare. This report presents a case of a STUMP in an older male patient, the origin of which could not be determined. The mainstay of treatment is surgical resection. Given this tumour type's uncertain biological potential and documented late recurrence in uterine STUMPs, prolonged surveillance is recommended.

Keywords: Smooth muscle tumours of uncertain malignant potential, STUMP, Leiomyoma, Leiomyosarcoma

INTRODUCTION

Smooth muscle tumours of uncertain malignant potential (STUMPs) are rare neoplasms that present diagnostic difficulties due to their intermediate morphology.[1] As their name suggests, STUMPs arise from smooth muscle cells and on histopathological review, demonstrate both benign and malignant features.[1] Specifically, to characterize a tumour as a STUMP, one or more of the following histological features must be present: tumour cell necrosis, cellular atypia, and an elevated mitotic index, with the value between that of a benign and a malignant tumour.[2] While these tumours can be locally invasive and fast growing, they seldom metastasis to distant structures.[3] STUMPs tend to develop in females more than males and are typically uterine in origin.[3,4] The symptomatology of STUMPs is dependent on the structure from which they arise, but often present with symptoms related to mass effect. For example, in large uterine STUMPs, presenting complaints can include pelvic pain, sensation of pelvic fullness or pressure, abnormal vaginal bleeding or urinary symptoms such as frequency or retention.[5] While these neoplasms have uncertain malignant potential, the mainstay treatment remains surgical resection.[5] While uterine STUMPs are uncommon, STUMPs arising from other structures, specifically in males, are significantly less frequent encountered.[4] This report will describe the case of an older male who presented with a pelvic mass with associated compressive symptoms, which was found to be a STUMP. Interestingly, the organ from which this lesion arose could not be definitively identified.

CASE REPORT

A 64-year-old male presented with an approximately six-month history of gradually worsening lower limb oedema, most prominent at night. Apart from fatigue, he did not report any other symptoms. At time of presentation, the patient did not have any significant past medical history and apart from a vasectomy twenty years prior, also did not have any surgical history. On examination, mobility was unencumbered and a large, firm, non-tender mass was palpable in the lower abdomen. He was also noted to have bilateral pitting oedema to his upper thighs and pelvis. On

investigation, renal function tests showed an eGFR of 49 ml/min and a raised creatinine to 132 µmol/l. Ca125 was raised to 41 but PSA, CEA and Ca19.9 were unremarkable. A computed tomography (CT) scan of his chest, abdomen and pelvis demonstrated an abdominopelvic mass measuring 27×17×15 cm in the craniocaudal, anteroposterior and transverse dimensions, respectively (Figure 1). The mass demonstrated areas of necrosis but no calcification. It was radiologically inseparable from the prostate and was producing significant mass effect on the distal ureters bilaterally with resultant severe bilateral hydronephrosis and hydroureter. Similarly, compression of the inferior vena cava was also noted. Subcutaneous oedema was reported in the lower abdominal wall and proximal thighs. There were enlarged inguinal lymph nodes with the largest on the right measuring 20×11 mm and the largest on the left measuring 17×22 mm. There were no other suspicious lesions or nodes on the remainder of the scan. A radiologically guided biopsy of the lesion was performed shortly thereafter with histopathological examination reporting findings of a soft tissue spindle cell lesion with bland morphology and no significant atypia or abnormal mitosis. A differential provided at this time was of a possible desmoid type fibromatosis.

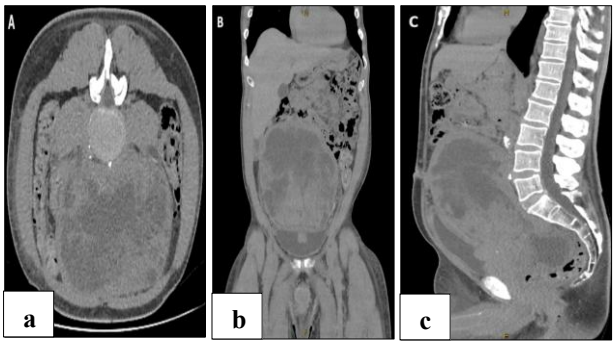

Figure 1: CT images demonstrating large, heterogenous pelvic mass – (a) axial, (b) coronal, and (c) sagittal.

Given the findings of a likely acute kidney injury and obstructed kidneys on imaging, the urology team was consulted, and the patient was planned for a cystoscopy and ureteric stenting. Intra-operatively, a digital rectal examination was performed. There was difficulty palpating the prostate, which appeared to be adherent to the abdominopelvic mass. Cystoscopy was then performed which demonstrated a long, occlusive prostate and there was difficulty visualizing the bladder neck due to the acute angle of the bladder. Using a wire, a flexible cystoscope was introduced however visualization and subsequent attempts at cannulating the ureteric openings were unsuccessful. Vision was further obstructed by bleeding from the prostate towards the end of the case; hence the procedure was aborted with percutaneous nephrostomies and anterograde stents placed a later date. Some days later, a laparotomy and resection of the pelvic mass along with a total cystoprostatectomy and formation of an ileal conduit was performed. The nephrostomies were later removed, and the post-operative course was otherwise unremarkable.

The resected specimen was sent for histopathologic review. It weighed 3318 g and measured 257×190×130 mm (Figure 2). Interestingly, while the tumour was attached to both the bladder muscle and the prostate, there was no evidence of infiltration into either structure. Microscopic examination revealed a lobulated mass comprised of fascicles of uniform spindle cells with mild cytologic atypia noted (Figure 3).

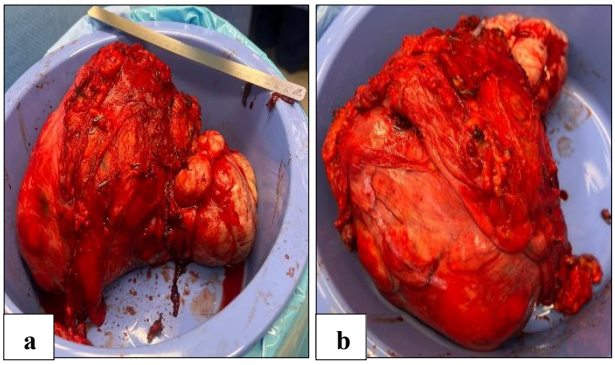

Figure 2 (a and b): Pelvic mass following resection, weighing 3318 g and measuring 257×190×130 mm.

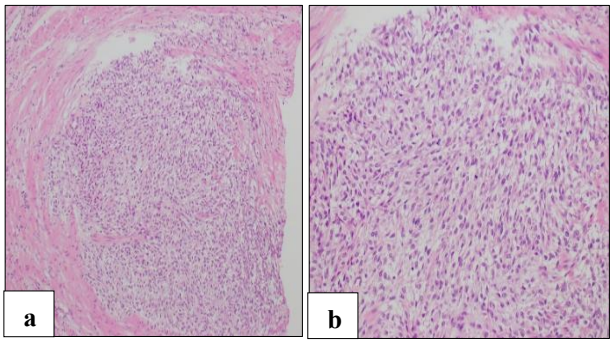

Figure 3 (a and b): Slides of different magnifications demonstrating spindle cells in fascicles, with one mitotic figure in view; cells are mostly uniform with mild nuclear atypia.

There was a focal area of two per 50 mitoses with the remainder of the lesion having a mitotic rate of less than one per 50 high power field (HPF). There were small foci of myxoid changes and fibrosis but no areas of necrosis and no infiltrative border. The mass was described as overall, having the features of a smooth muscle tumour arising from the abdominopelvic cavity. While majority of the mass did not show any cytologic atypia, the presence of focal mild atypia with a focal mitotic rate of 2 per 50 HPF and the results of the immunohistochemical profile (Table 1) performed allowed for it to be classified as a STUMP.

Table 1: Immunohistochemical profile of the resected lesion.

Characteristics	Immunohistochemical profile
Positive	Desmin, SMA, Vimentin, ER (patchy), AR (patchy) and PR (patchy)
Negative	AE1/3, CAM5.2, EMA, HMB45, SOX10, NKX3.1, STAT6, DOG1, MelanA, β-catenin (negative nuclear staining), Calretinin, MUC4, Cyclin D1, CD31, CD34, CD117, S100 and ALK

Given the histopathological findings, the patient did not undergo any local radiotherapy or systemic chemotherapy, however, was closely monitored with initially six monthly then yearly imaging, which remains ongoing. CT and positron emission tomography (PET) imaging has not identified any local or distant recurrence of disease at time of publishing.

DISCUSSION

STUMPs are rare neoplasms situated along a biological continuum between benign leiomyomas and leiomyosarcomas.[6] Although well recognised in the uterus, their occurrence in males is exceedingly uncommon and extra-genitourinary presentations, particularly those arising within the abdominopelvic cavity without a clear organ of origin, are exceptional.[1,4] The present case contributes to the limited but growing literature demonstrating that STUMPs can arise in virtually any location containing smooth muscle or its mesenchymal precursors.

Diagnosis in males is particularly challenging because these tumours do not contain organ-specific markers and share histological features with several mimics. Key differentials include leiomyoma, leiomyosarcoma, gastrointestinal stromal tumours, desmoid-type fibromatosis, and other soft tissue sarcomas.[7] Immunohistochemistry was therefore essential in ruling out these other tumour types. Strong desmin, SMA, and vimentin positivity confirmed smooth muscle differentiation, while negativity for KIT (CD117), DOG1, and CD34 excluded GIST.7 Absence of epithelial markers (AE1/3, CAM5.2, EMA), neural and PEComa markers (S100, SOX10, MelanA, HMB45), and lineage-specific markers such as β-catenin, NKX3.1, STAT6, MUC4, Cyclin D1, and ALK effectively ruled out other major diagnostic possibilities, such as metastatic prostate carcinoma or solitary fibrous tumour.[7] Patchy oestrogen receptor, progesterone receptor, and androgen receptor expression, although classically associated with uterine smooth muscle tumours, is well documented in extra-uterine smooth muscle neoplasms and in this setting represents a non-specific Müllerian-type immune-phenotype rather than Müllerian origin.[8] These markers provide supportive evidence of smooth muscle biology and fall within the documented immunophenotypic spectrum of extra-uterine STUMPs.

The biological behaviour of extra-uterine STUMPs remains poorly characterized. Available evidence on STUMP outcomes is limited, with variable recurrence and rare malignant transformation reported in uterine cases, suggesting that extra-uterine examples, although seldom reported, may behave in a similarly unpredictable fashion.[3,9] Tumours in deep anatomic compartments, such as the retroperitoneum or abdominopelvic cavity, may present late, as seen in our case, and pose challenges for achieving complete resection, potentially elevating recurrence risk. Although the mass in our patient was removed with negative margins, this does not eliminate the possibility of local recurrence.

Surgical excision with clear margins is the mainstay of treatment.[1,5,9] There is no established role for adjuvant therapy, largely owing to the scarcity of data and absence of validated risk-stratification systems.[3] Most authors of published case reports adopted an observation approach rather than administering adjuvant chemotherapy or radiotherapy, reserving additional treatment for recurrent or overtly malignant transformation.[1,4] This evidence gap is particularly pronounced in extra-uterine cases in males, for which guidance is almost entirely derived from isolated case reports.

Given the tumour's unpredictable natural history, long-term surveillance is recommended.[3,10] A reasonable follow-up strategy parallels protocols for low-grade soft tissue sarcomas, incorporating cross-sectional imaging every 6–12 months for several years, followed by annual imaging thereafter.[3,6,10]

Reports of very late recurrences sometimes more than a decade after initial excision in uterine STUMPs may support the implementation of an extended period of monitoring.[3]

CONCLUSION

This case highlights the rarity of extra-uterine STUMPs in male patients and underscores the diagnostic challenges posed by their non-specific clinical and histological features. Comprehensive immunohistochemistry is essential to exclude a broad range of mimics and to confirm true smooth muscle differentiation. Although completely excised, the tumour's uncertain biological potential and the precedent of late recurrence in analogous uterine lesions necessitate prolonged surveillance. This report adds to the limited literature documenting extra-uterine STUMPs in males and reinforces the need for continued recognition, careful diagnostic evaluation, and long-term follow-up in such cases.

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