Solitary Fibrous Tumor of Penile Root: First Case in Oman

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Abstract

Background: Solitary fibrous tumor (SFT) is a rare spindle-cell mesenchymal tumor of myofibroblastic origin. Penile SFT has been reported in the literature in four separate case reports only. We present the first case of penile SFT in Oman, and the fourth case in the literature. SFT presents as a slow growing, often asymptomatic mass, generally affecting middle-aged adult males. Case presentation: A 37-year-old man presented with a small and gradually growing lesion at the base of his penis for a duration of 4 months. On physical examination; a well-defined, nodular non-fluctuant, non-tender, painless, solid mass, measuring 4x3 cm was palpated at the root of the right side of the penis. Magnetic resonance imaging showed 4x2.5 cm intensely enhancing heterogenous mass lesion arising from the soft tissue of right inferio-lateral root of penis. Surgical excision was performed through a perineal incision. Pathologically the features were consistent with Solitary Fibrous Tumour (SFT) of the penis. Conclusions: SFT rarely occurs in genital tract and penile presentation is among the rarest. The mass is usually well circumscribed, painless and slowly growing in nature. After differential diagnosis, surgical excision is mandatory to rule out malignant lesions like Fibrosarcoma. Although SFTs are benign in nature, close follow up is recommended due to possibility of local or distant recurrences.

Keywords: Fibrous; Tumour; Penile; Oman.

Introduction

Solitary fibrous tumor (SFT) is a rare spindle-cell mesenchymal tumor of myofibroblastic origin. It has an intermediate behavioral pattern which rarely metastasizes. This tumor was described by Muray in 1942 as a pleural based lesion. ¹ Thirty percent of SFT occurs in sites other than pleura (extrapleural). Deep muscles of proximal extremity, head, neck and retroperitoneum are common locations for extrapleural SFT. ² Penile SFT has been reported in the literature in only four separate case reports which were published in 2015, ³ 2017, ⁴ 2019, ⁵ and 2023. ⁶ We present the first case of penile SFT in Oman, and the fourth case in the literature.

Case Report

A 37-year-old man presented to our clinic with a small and gradually growing lesion at the base of his penis for a duration of 4 months. On physical examination; a well-defined, nodular non-fluctuant, non-tender, painless, solid mass, measuring 4x3 cm was palpated at the root of the right side of the penis. No palpable lymph nodes were detected. No abnormalities were detected on laboratory analysis including testicular tumor markers. Magnetic resonance imaging showed 4x2.5 cm intensely enhancing heterogenous mass lesion arising from the soft tissue of right inferiolateral root of penis, abutting the tunica albuginea but not invading it or the cord. Decision of surgical excision through perineal incision under general anesthesia was performed. During surgical exploration, it was observed that the mass had clear boundaries with an evident capsule. It originated deep from the root of penis and close to the right corpus

cavernosum. The mass was not in close contact with the spermatic cord or the urethra. Blind and sharp dissections were performed around the mass and the mass was sharply excised after ligation of a basal vascular stalk [Figure 1]. Patient was discharged on the first postoperative day, uneventfully. On multi-disciplinary meeting the decision was made to follow up the patient clinically every 6 months and do imaging as needed.

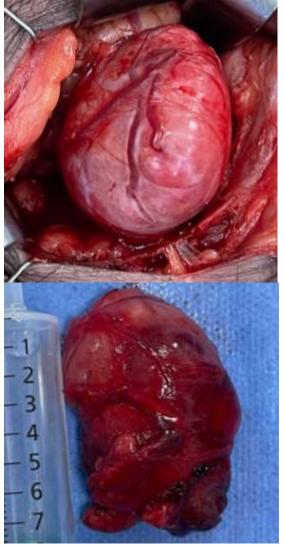


Figure 1: Gross features of the lesion specimen are shown. A: mass before excision; B: mass after excision

Macroscopically, the resected penile mass measured 5x3x1.4cm. It consisted of an encapsulated nodular tumour with smooth external surface showing congested blood vessels. Cut surface of tumour showed heterogeneous yellow, white and hemorrhagic areas. Histological sections from the penile mass showed an encapsulated well circumscribed spindle cell neoplasm, composed of hypercellular patternless sheets of fairly uniform oval to spindle cells set in highly vascular and variable scant fibrocollagenous stroma. Occasional thick short fibrous bands are noted and there are hypocellular tumour zones seen particularly at the periphery with the stroma appearing markedly edematous, partly myxoid and rich in dispersed mast cells. The tumour cells have elongated plump vesicular nuclei, inconspicuous nucleoli and scant eosinophilic cytoplasm. There is low mitotic activity observed; ranging from 2 up to 3 mitosis /10 HPF. There is no necrosis or significant cytological atypia identified. The neoplastic cells surround variably sized thin and thick walled blood vessels, some of which are large ectatic with irregular branching walls "staghorn-like" and others are small capillary sized appearing focally arborizing. There is no tumour infiltration of surrounding attached fibrofatty tissue. The spindle tumour cells show diffuse strong positive staining for STAT-6, CD99 and BCL2. CD34 is focally positive in some tumour cells and also highlights many blood vessels. The spindle tumour cells are negative

for S100, Desmin, SMA, CD31, D2-40 and HHV8, and Calponin. The features are consistent with Solitary Fibrous Tumour (SFT) [Figure 2].

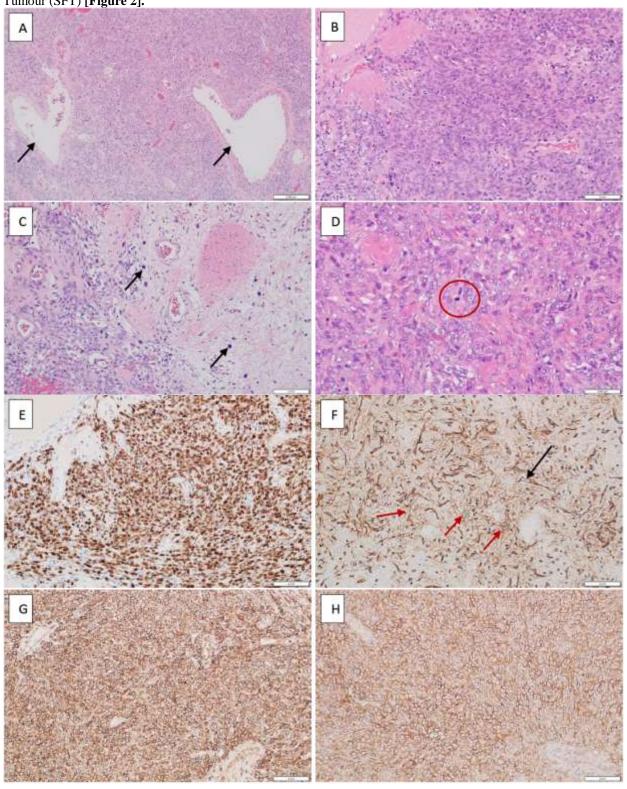


Figure 2: (A) Hematoxylin & Eosin (H&E) stained section of the penile mass showing spindle cell neoplasm with prominent large ectatic branching "staghorn-like" blood vessels (arrows) [magnification x100]; (B) H&E stain

showing tumour consisting of hypercellular patternless sheets of fairly uniform oval to spindle cells [magnification x200]; (C) H&E stained section of tumour showing focal hypocellular edematous partly myxoid areas rich in scattered mast cells (arrows) [magnification x200]; (D) H&E stained section showing a mitotic figure (circle) [magnification x400]; (E) STAT6 immunohistochemical stain reveals diffuse strong positive nuclear staining in tumour cells [magnification x200]; (F) CD34 immunohistochemical stain is focally positive in some spindle tumour cells (red arrows) and also stains blood vessels (black arrow) [magnification x200]; (G) & (H) BCL2 and CD99 immunohistochemical stains are diffusely positive in tumour cells; respectively [magnification x200].

Discussion

Solitary fibrous tumors (SFT) are rare mesenchymal neoplasms of fibroblastic origin representing less than 2% of all soft-tissue tumors. They manifest as a slow growing, often asymptomatic mass which affects middle-aged adults. Clinical presentation of patients depends on location of the tumor. Tumors occurring in the abdomen or pelvis give symptoms due to the compression of the neighboring organs. Spinal tumors present with localized pain due to spinal cord compression or a palpable mass. Abdominal and pelvic tumors presented with abdominal or back pain. Prostate tumors presented with urinary retention and constipation, or voiding difficulties, hematuria, incidental imaging findings and lower abdominal discomfort. Penile tumor present with a slow growing painless mass. In contrary malignant penile lesions like Fibrosarcoma of the corpus cavernosum are characterized by pain, enlargement, penile erection and urinary tract obstruction.

In the literature, there is no specific laboratory test available for the diagnosis of SFT. However, 10% of patients have hypoglycemia due to increased production of Insulin like Growth Factor 2. ¹⁴ Differentiation between SFT and other solid tumors is also not possible using radiological studies. However, on CT, SFT appears as as homogeneous mass with well-circumscribed margins, and without any sign of invasion. On the other hand, variety of enhancement, necrosis and hemorrhage have been demonstrated in SFT making limitations for the differential diagnosis. ^{8,10} Hence, tumor sampling via needle biopsy is a useful tool for the differential diagnosis of SFT, although biopsy of paratesticular and spermatic cord masses is not recommended due to the possibility of spreading the cancer cells. ¹² It has been suggested that close follow up for at least five years for patients who had received a treatment for SFT in oral cavity, deep extremity, female genital tract, but a standard follow- up proposal is not available. ¹⁵ Individual follow-up strategies usually include radiological studies and physical examination. ⁷ The treatment of choice is surgical resection, however, disease recurrence or progression to malignant tumors can occur in 10%–30% of patients. ¹⁶ Multiple risk factors and risk stratification systems have been investigated to predict which patients are at risk of recurrence but the most significant factors found were high mitotic index, necrosis and Ki67 index. ¹⁶

Conclusion

SFT rarely occurs in genital tract and penile presentation is among the rarest. The mass is usually well circumscribed, painless and slowly growing in nature. After differential diagnosis, surgical excision is mandatory to rule out malignant lesions like Fibrosarcoma. Although SFTs are benign in nature, close follow up is recommended due to possibility of local or distant recurrences. Further studies with larger number of cases are required to better stratify the outcome of patients with SFT in this particular anatomical location.

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