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Malignant solitary fibrous tumor of the penis: A case report of rare malignancy

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ABSTRACT

Malignant Solitary fibrous tumor (SFT) is a rare mesenchymal tumor that consists of spindle cell components. The occurrence of SFT in the genitourinary tract is extremely rare. So, there is no clear algorithm for the management of this case. We report A 33-year-old male who complained of recurrent penile swelling in the last 7 months after underwent surgery 3 months earlier. The tumor re-enlarged from the surgical wound's previous sutures. Total penectomy was performed followed by bilateral inguinal lymphadenectomy. Perineostomy was performed for urinary diversion. Long-term follow-up after surgery is suggested due to the risk of recurrences and metastasis.

1. Introduction

Malignant Solitary fibrous tumor (SFT) is a rare mesenchymal tumor that consists of spindle cell components arranged in a patternless distribution, and difficult to diagnose due to difficulty distinguishing to other benign or malignant spindle cell mesenchymal tumors.[1] Confirmation from the immunohistochemistry (IHC) examination is required.[2]

This tumor can appear anywhere in the body, but the occurrence of SFT in the genitourinary tract is extremely rare. There are very few reported cases of SFT of the penis. To the best of our knowledge, we only found two reports mentioning the incidence of SFT of the penis. Due to its rarity, there is no exact algorithm for managing this case.

We reported a patient with a bulky mass, irregular swelling, and ulcerative nodules of the penis, which was recurrent four months after the first resection.

2. Case presentation

A 33-year-old married and fertile male complained of swelling in the last seven months after undergoing tumor resection on his penile shaft three months earlier. After the first surgery, the patient reported that the tumor had re-enlarged from the previous surgical wound.

He had previously a history of penile trauma at eight years old and developed 0.5 × 0.5 mm lump on his subcoronal penis. The lump's size remained constant until he was sexually active nine years ago. The patient had no other history of the previous disease.

On genital examination, there was a bulky mass with irregular swelling and fleshy nodule mass occupying the region of the penis (Fig. 1). Regional lymphadenopathy was not palpable.

A contrast computed tomography (CT) scan of the abdomen and pelvis reveals well-defined, partially irregular edges, and hypodense mass occupying the region of the penis measuring 13.4 x 6.4 × 12.9 cm (Fig. 2a). Right and left inguinal lymphadenopathy with a diameter of 0.7–1 cm was also seen (Fig. 2b). There were no signs of distant metastases. Total penectomy with bilateral inguinal lymphadenectomy and perineostomy was performed. We also conduct frozen sections of the penile corpus cavernosum to find any positive margin. Tumor-free margin was found.

Histopathology examination after surgery revealed spindle cell malignancy of the penis (Fig. 3) infiltrating the corpus cavernosum (pT3), but no lymph node involvement was found (pN0). Immunohistochemistry examination confirmed intense positivity of CD34 and vimentin as markers of a malignant SFT of the penis. The patient was then decided to receive adjuvant chemotherapy with six cycles AIM (doxorubicin,

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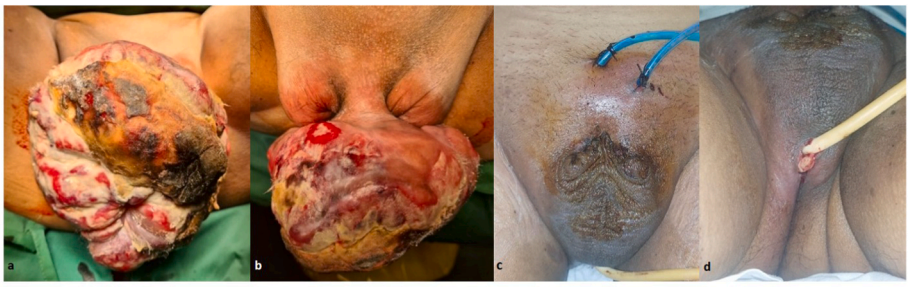

Fig. 1. (a,b) Recurrent tumor growth after first surgical management reveals a bulky mass and alters the normal shape of the penis.; **(c,d)** Postoperative picture after total penectomy, perineostomy, and bilateral inguinal lymph node dissection. Urinary catheter was placed postoperatively at the perineostomy site.

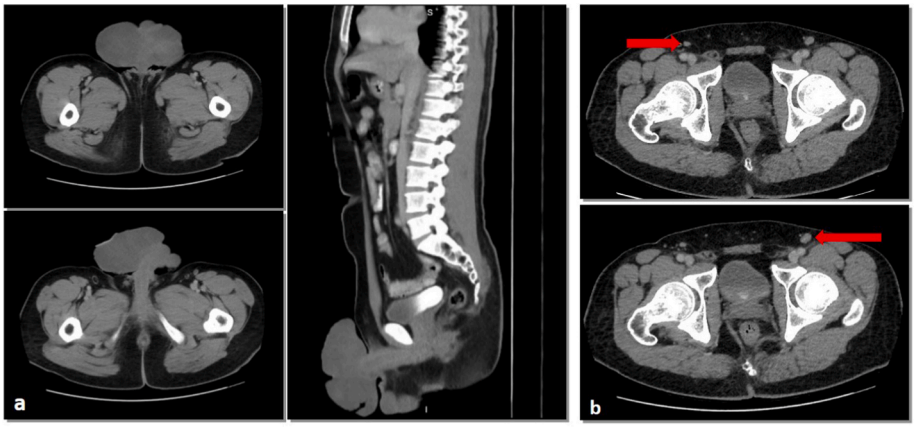

Fig. 2. a). Abdominal and Pelvic CT (axial and sagittal) shows tumor mass occupying the region of the penis; **b).** Right and left inguinal lymphadenopathy.

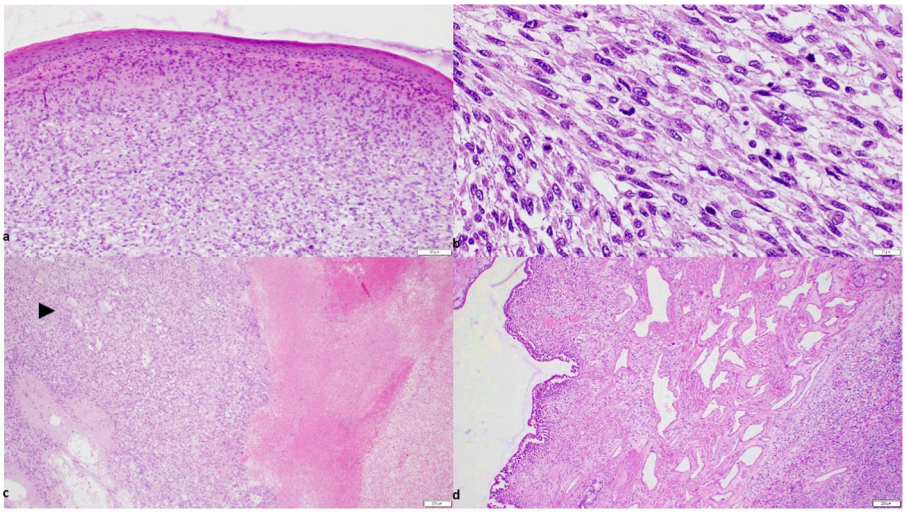

Fig. 3. a). Sections showed tumor located adjacent to surface epithelia. Tumor arranged in so-called "patternless" fashion. No apparent fascicles. (H&E x10); **b).** Tumor composed of spindle cells, moderate to high nuclear pleomorphism was shown. Tumor cells showed coarse chromatin, some with prominent nucleoli. Mitosis was observed; **c).** Tumor showed cystic degeneration (arrowhead), and area of necrosis (right side) (H&E x4); **d).** Tumor invaded the cavernous body (H&E x4).

ifosfamide, mesna) regimen and radiotherapy from a multidisciplinary meeting.

3. Discussion

Malignant SFT is a rare soft tissue malignancy consisting of spindle cells. It can occur anywhere in the body, most commonly in the pleura, head, and neck, but extremely rare in the genitourinary tract. According to other studies, malignant SFTs can develop from an already-existing benign SFT; meanwhile primary malignant SFT is an extremely rare event.[1] To the best of our knowledge, we only discovered two reports that report SFT of the penis, but had a different clinical picture and did not show destructive features as found in our case.

In this case report, the patient presented with a bulky mass with irregular swelling and nodules occupying the region of the penis. Misdiagnosis occurs due to the difficulties in diagnosing SFT since its morphology is unspecific and patternless. Histopathologic examination of SFT in our patient revealed spindle cells shaped with oval nuclei.

Therefore, immunohistochemistry (IHC) analysis is required for confirmation.[2] From two case reports of SFT of the penis, CD34 has been identified as one of the most consistent conventional markers. Cubuk et al. added CD99 and bcl2 markers which also revealed diffuse and strong positivity.[3,4] Similarly, immunohistochemical staining on CD34 and vimentin revealed a diffuse positive result in our case.

The etiology of SFT was not clear. The last report from Castellani and Cubuk does not reveal the medical and surgical history of SFT of the penis.[3,4] No records of promiscuity were found in our case and other case reports. Meanwhile, our patient reported prior trauma when he was eight years old. Scars in the form of small lumps resembling keloids become the origin of tumor enlargement.

Malignant SFT reported has a wide age range. Siregar et al. reported that SFT ranges in age from 14 to 68 years, with a median age of 39 years.[2] Castellani et al. reported that their patient was a 44-year-old man, meanwhile, Cubuk et al. reported a 55-year-old man complaining of a small, gradually growing lesion on his penis.[3,4] In our case, the patient reported the first complaint when he was eight years old. Then, the tumor enlarged slowly after his marriage nine years ago. The patient gets his first resection of the penile tumor in another hospital. Following the initial surgical excision, the tumor developed quickly from the wound, causing pain and disrupting normal activities. According to other studies, the recurrence rate for malignant SFT ranges from 14% to 86%, with 18% evidence of distant metastasis.[1] However, there is no local recurrence nor distant metastasis found from the latest report of penile SFT.[3,4]

There is no agreement on how to treat this disease. Complete surgical resection with free margins is the recommended treatment for localized malignant SFTs.[1] In cases where the tumor is unresectable, radiotherapy and adjuvant chemotherapy have been considered, although there is no proof of significant benefits.[1] In our case, we performed total penectomy, bilateral inguinal lymphadenectomy, and perineostomy. After surgical management, our patient is scheduled for adjuvant chemotherapy and radiotherapy, based on NCCN Guidelines which state that adjuvant therapy is spesifically considered to be given to infiltrative histologies regardless tumor margins.[5]

4. Conclusions

Malignant SFT in the penis is an extremely rare case. Our case showed local aggressive features without evidence of lymph node and other organ metastasis. However, since it has a risk of recurrence and metastasis, we planned to continue the treatment with adjuvant chemotherapy and radiotherapy. Moreover, long-term follow-up after surgery is mandatory.

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