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## LETTER TO THE EDITOR

Male Health

# Superficial angiomyxoma of penis: a case report of a 6-year follow-up

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Dear Editor,

We report a case of an 18-year-old Chinese man with two recurring asymptomatic superficial angiomyxomas on his penis. After an incisional biopsy, we resected the masses on the proximal and distal ends of the penis. Clinical and pathological findings contributed to the diagnosis of penial superficial angiomyxoma. An 80-month follow-up showed no sign of recurrence, and the enlargement of bilateral inguinal lymph nodes disappeared 8 months after complete resection of the tumors. Angiomyxoma is relatively rare in soft tissue tumors,<sup>1</sup> and usually includes three types: aggressive, superficial, and angiomyofibroblastoma.<sup>2</sup> To the best of our knowledge, this is the first case of a recurring penial superficial angiomyxoma (SAM).

An 18-year-old man was referred to our hospital on December 12, 2008, with two asymptomatic subcutaneous masses on the proximal and distal ends of his penis. His medical history indicated that he underwent local excision of a small nodular lesion on the proximal end of his penis at a medical clinic in 2006 without an excisional biopsy after the operation. Six months after the local excision, the subcutaneous mass recurred, and a new subcutaneous mass formed on the distal end of his penis. The initial physical examination showed that the lesions were soft, moderately firm, nontender, and subcutaneous masses (**Figure 1**). CT scan showed bilateral inguinal lymph nodes enlargement. An incisional biopsy before surgical excision revealed that the masses were benign tumors. The patient underwent complete resection of the tumors. On gross examination, the resected masses on the proximal and distal ends of the penis were 5 cm × 4 cm × 3.5 cm and 4 cm × 3 cm × 3 cm, respectively. The tumors were grayish-white, and the cut surface had a yellow white, gelatinous, glistening appearance, and multiple-sized pseudocysts (**Figure 2**).

Histopathological analysis showed that the background comprised of a diffuse collagenous myxoid, and spindle- or stellate-shaped mesenchymal cells were scattered throughout the tissue. Thick- and thin-walled, small to medium-sized, and hyaline degenerated blood vessels were diffusely distributed throughout the stroma in a disordered arrangement. A diffuse infiltrate of neutrophils was present in the stroma (**Figure 3**). Immunohistochemical test revealed that the

tumor cells were diffusely positive for vimentin, CD34, and CD44, but negative for smooth muscle actin, desmin, S-100 protein, and pan-keratins (**Figure 4**). Clinical and pathological findings contributed to the diagnosis of penial superficial angiomyxoma. The margins of the sections were free from tumor cells.

An 80-month follow-up showed no sign of SAM recurrence, and the enlargement of bilateral inguinal lymph nodes disappeared 8 months after complete resection of the tumors.

Superficial angiomyxoma is a rare, benign cutaneous tumor, which was first described in 1988 by Allen *et al.*<sup>3</sup> Superficial angiomyxoma has a tendency for local recurrence after incomplete excision, but lacks metastatic potential.<sup>3,4</sup> The tumor usually presents as a nodular lesion on the trunk, head, neck, and even female genital region such as vulva. There have been only six reported cases of superficial angiomyxoma on the scrotum.<sup>1,4,5</sup>

Superficial angiomyxoma differs from aggressive angiomyxoma,<sup>4,6</sup> and rarely occurs in the genital region in males.<sup>1,4,6</sup> In this unique case, an 18-year-old male, was referred to our hospital because of recurring masses on his penis. His clinical, histopathological, and immunohistochemical findings showed a polypoid tumor without invasive growth, and no apparent atypism in the tumor cells. Based on the above factors, aggressive angiomyxoma and other malignant tumors were ruled out. The regression of enlarged inguinal lymph nodes after tumor resection demonstrated its benign feature. Neurogenic tumors such as dermal nerve sheath myxoma and myxoid neurofibroma were unlikely, since none of the tumor cells expressed S-100 protein.<sup>6</sup> Superficial angiomyxomas are generally immunoreactive with vimentin and CD34,<sup>2</sup> which was consistent with the present case. In addition, neutrophilic infiltration in superficial angiomyxoma distinguishes it from other myxoid lesions, and is an important histological clue for the differential diagnosis.<sup>4,6</sup> Complete surgical excision is very crucial for superficial angiomyxomas because of its high rate of local recurrence between 20% and 40%.<sup>2,7,8</sup>

In this letter, we describe a complete surgical excision and long-term follow-up of penial superficial angiomyxomas. In the present case, no recurrence was observed during a 6-year follow-up, due to complete surgical excision. Though superficial angiomyxomas have a good overall prognosis, without invasion of deeper structures,<sup>2,7,9</sup> the patients should be followed up for long-term.

### AUTHOR CONTRIBUTIONS

YCW and GPZ carried out the surgery and follow-up; YCW and ZX drafted the manuscript; XML carried out histopathological analysis and immunohistochemical testing; ZPW participated in the design

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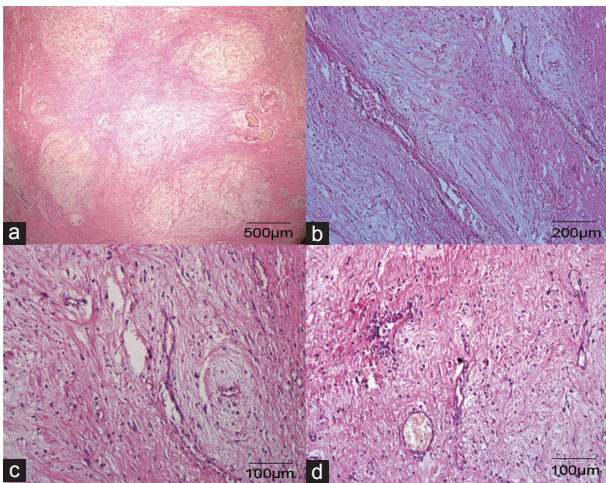
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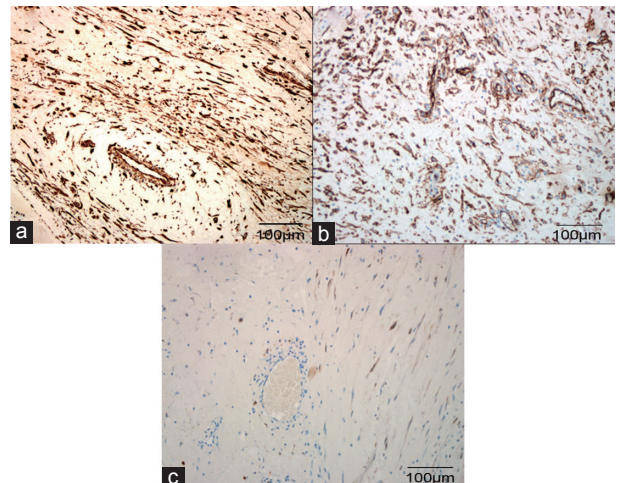
**Figure 1:** The two recurrent asymptomatic subcutaneous masses were on the proximal end and the distal end of the penis.



**Figure 2:** The cut surface had a yellow white, gelatinous, glistening appearance, and multiple-sized pseudocysts.



**Figure 3:** (a) Superficial angiomyxoma (SAM) consists of a conglomerate of moderately-to-sparsely cellular angiomyxoid lobules (scale bar = 500  $\mu\text{m}$ ). (b) Each lobule consists of spindle-shaped or stellate tumor cells diffusely distributed in a myxoid stroma (scale bar = 200  $\mu\text{m}$ ). (c) Small-to-medium-sized blood vessels were scattered throughout the stroma (scale bar = 100  $\mu\text{m}$ ). (d) A diffuse infiltrate of neutrophils is present in the stroma (scale bar = 100  $\mu\text{m}$ ).



**Figure 4:** Immunohistochemical staining revealed that the tumor cells constantly expressed. (a) Vimentin, (b) CD34, and (c) S-100 occasionally expressed stroma (scale bar = 100  $\mu\text{m}$ ).

of the study and helped to draft the manuscript. All authors read and approved the final manuscript.

#### COMPETING INTERESTS

None of the authors declare competing financial interests.

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