Case Report

Superficial angiomyxoma on the scrotum

陰囊上的淺表性血管黏液瘤

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Superficial angiomyxoma (SAM) is a rare benign multilobulated cutaneous tumour, comprised of a prominent myxoid matrix and numerous blood vessels. Scrotal SAM is extremely rare. We report a case of SAM on the left scrotum and the tumour was completely excised.

淺表性血管粘液瘤是一種罕見的多發小葉狀良性皮膚腫瘤，由顯著的粘液樣基質和許多血管組成。陰囊淺表性血管粘液瘤更是極度罕見，我們報告一例左側陰囊的淺表性血管粘液瘤病例，腫瘤被完全切除。

**Keywords:** Excision, benign, scrotum, superficial angiomyxoma, tumour

關鍵詞：切除、良性、陰囊、淺表血管黏液瘤、腫瘤

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**Introduction**

Superficial angiomyxoma (SAM) is a rare benign multilobulated cutaneous tumour, comprising of a prominent myxoid matrix and numerous blood vessels. SAM is more common in middle-aged men than in women and is usually located on the trunk, head and neck or lower extremities. Scrotal SAM is extremely rare. Herein, we report a case of SAM on the left scrotum of a 27-year-old male and review the literature.

**Case report**

A 27-year-old Chinese male presented to our clinic with an enlarging cutaneous mass on the left scrotum. The tumour had grown gradually for three years. The patient had no local pain, fever, or
urinary tract symptoms; nor did he have any history of local injury. The patient's past medical and family history were normal. Clinical examination revealed a moderate-hard, flesh-coloured, non-transparent mass, measuring 2.7x2.3x1.5 cm, on the left scrotum (Figure 1). The mass had a smooth surface and multi-lobulated nodular features with dilated blood vessels on the surface. The tumour was completely excised under local anaesthesia. A chest X-ray and a cardiac colour ultrasound showed no abnormalities. Histopathologically, the specimen showed a well-circumscribed, multilobular myxoid lesion in the dermis beneath the normal epidermis, proliferation of thin-walled vessels and a sparsely cellular proliferation of satellite and spindle-shaped cells deposited in an abundant myxoid matrix (Figure 2a). The fibroblast-like cells of the tumour did not exhibit cytological atypia or mitotic figures (Figure 2b). Presence of a neutrophilic infiltrate could be seen. The tumour was immunoreactive for vimentin, CD34 and actin (HHF35) (Figures 3a, b, c). No immunoreactivity was present for progesterone receptor (PR), oestrogen receptor (ER), androgen receptor (AR), desmin and S-100 protein. A cytogenetic karyotypes analysis was performed on peripheral blood and no abnormal chromosomal complement was observed. Based on the clinical and histological findings, the patient was diagnosed as superficial angiomyxoma. The margins of the sections were free of tumour cells. There was no recurrence 8 months after excision and he is currently still on follow-up.

Figure 1. Clinical examination revealed a firm, flesh-coloured, mass on the left scrotum.

Figure 2. Histopathological findings. (a) Photomicrograph shows proliferation of thin-walled vessels and a sparse cellular proliferation of satellite and spindle-shaped cells deposited in an abundant myxoid matrix (Haematoxylin-eosin stain, original magnification x40). (b) High-powered photomicrograph shows no cytological atypia and atypical mitosis (Haematoxylin-eosin stain, original magnification x100).
Superficial angiomyxoma on the scrotum

Discussion

SAM is a rare benign cutaneous tumour, which was first described in 1988 by Allen et al.\(^1\) SAM has a tendency for local recurrence after incomplete excision, but lacks metastatic potential. The prevalence of SAM is very low (approximately 0.0008-0.3%), shows slight male predilection, and usually presents between 20-40 years of age.\(^2\) SAM usually occurs on the head and neck, trunk, and extremities. To the present authors' knowledge, there have been only nine reported cases of SAM on the scrotum. Clinical findings of previously reported cases and our case are summarised in Table 1. Immunohistochemically, the tumour cells stained for ER and PR in more than 90% of aggressive angiomyxoma (AAM) female tissues. There also have been few reports on the expression of these sex hormone receptors in AAM males.\(^3,4\) In our immunostaining studies, the tumour cells of SAM did not express ER, AR and PR. SAM rarely occurs in the genital region in males. SAM is generally immunoreactive with vimentin and CD34, which was consistent with the present case. Immunohistochemistry showed that the tumour cells were also positive for actin (HHF35) in our case. His clinical, histopathological, and immunohistochemical findings showed a large polypoid tumour no invasive component, and no apparent atypia in the tumour cells. Based on the above findings, AAM and other malignant tumours were ruled out. There have been several reports on the abnormal cytogenetic findings of AAM.\(^5\) A cytogenetic karyotypes analysis was performed on peripheral blood of our case, but no abnormal chromosomal complement was observed. SAM has a good overall prognosis, without invasion of deeper structures. Careful follow-up is required, since a SAM can be an early and only manifestation of Carney complex and the tumour can recur locally after excision in 30-40% of patients with incomplete resection.\(^1\)
Table 1. Clinical findings of previously reported cases and our case

<table>
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<tr>
<th>Reference</th>
<th>Age (year)</th>
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<th>Size (cm)</th>
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<tr>
<td>1997, Fetsch JF et al.</td>
<td>55</td>
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<td>3.5</td>
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<td>1999, Calonje E et al.</td>
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<td>Scrotum</td>
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<td>2011, Nakamura M and Tokura Y</td>
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<tr>
<td>2013, Jung HJ and Kim DY</td>
<td>69</td>
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<td>3.0x2.8x2.2</td>
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<td>2015, Wang Z et al.</td>
<td>25</td>
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<td>6.0x2.0</td>
<td>China</td>
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<td>2015, Lee CU et al.</td>
<td>56</td>
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<td>2017, Current case</td>
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<td>Left scrotum</td>
<td>2.7x2.3x1.5</td>
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References