



Oncology

Angiomyomatous Leiomyoma of a Female Urethral Meatus Recurrence After Seven Years of the Resection: A Case Report



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ABSTRACT

A 20 years old woman had an external urethral orifice mass and received an excision operation. Seven years later, she complained a tumor with pain that was similar to the previous tumor. She underwent the tumor removal. Pathological diagnosis was a urethral angiomyomatous leiomyoma in the new concept of estrogen receptor-positive smooth muscle tumors.

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Introduction

Leiomyomas are rarely encountered in the genitourinary tract outside the uterus.^{1,2} Leiomyoma is a benign smooth muscle tumor which is rarely found in urethra. It often appears in females during their reproductive age. There are few reported cases of tumor recurrence treated by a repeat excision in the literature to date.³

A case report

A 20 years old woman had an external urethral orifice mass and received an excision operation at Takaki Clinic in 2009. Pathological findings were urethral leiomyoma with benign nature at that time. She complained a tumor with pain that was similar as the tumor removed before (Fig. 1). She was introduced Uonuma Institute of Community Medicine, Niigata University Medical and Dental Hospital in October, 2015. She underwent cystoscopic examination and had no abnormal findings in urethra and urinary bladder. Pelvic CT and MRI revealed polypoid lesions with a contrast effect on the external urethral orifice at the size of 2.5 cm in diameter (Fig. 2). She underwent the tumor removal in January, 2016.



Figure 1. Gross appearance. The tumor is on the external urethral orifice.

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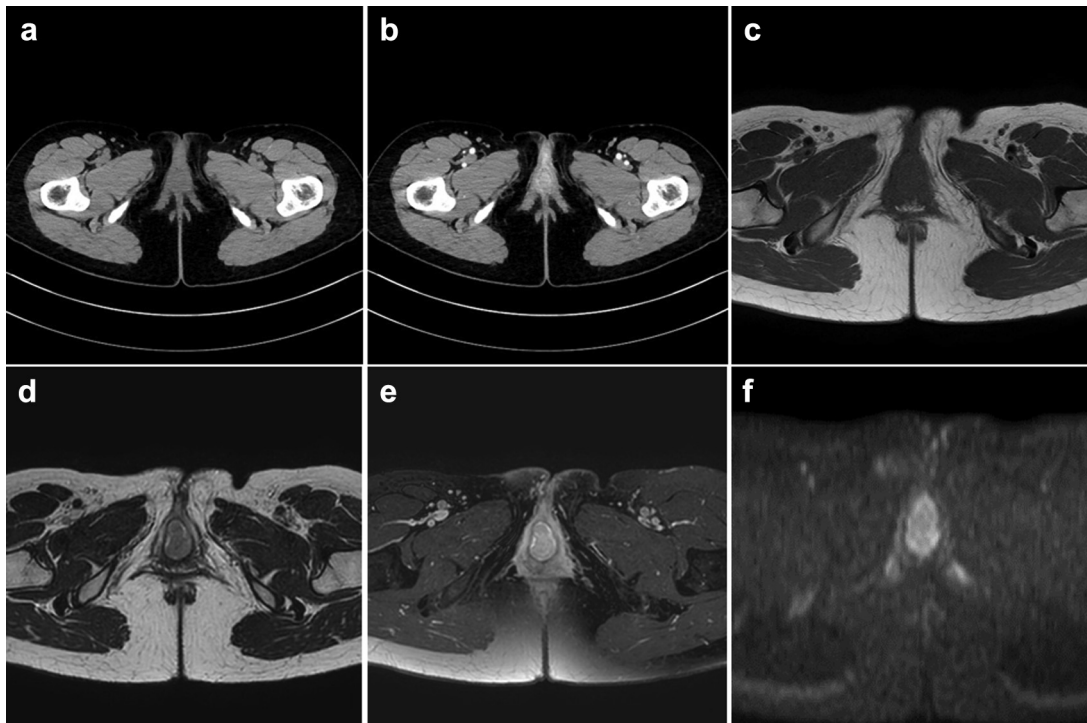


Figure 2. Pelvic CT and MRI findings. CT appearances. A Soft tissue density mass with homogenous contrast enhancement is seen at the external urethral meatus (a,b). MRI appearances. On a T1-weighted axial image, the mass shows intermediate signal intensity (c). On T2-weighted image and diffusion-weighted image, the lesion demonstrates slightly hyper-intensity (d,e). Homogenous contrast enhancement is observed (f).

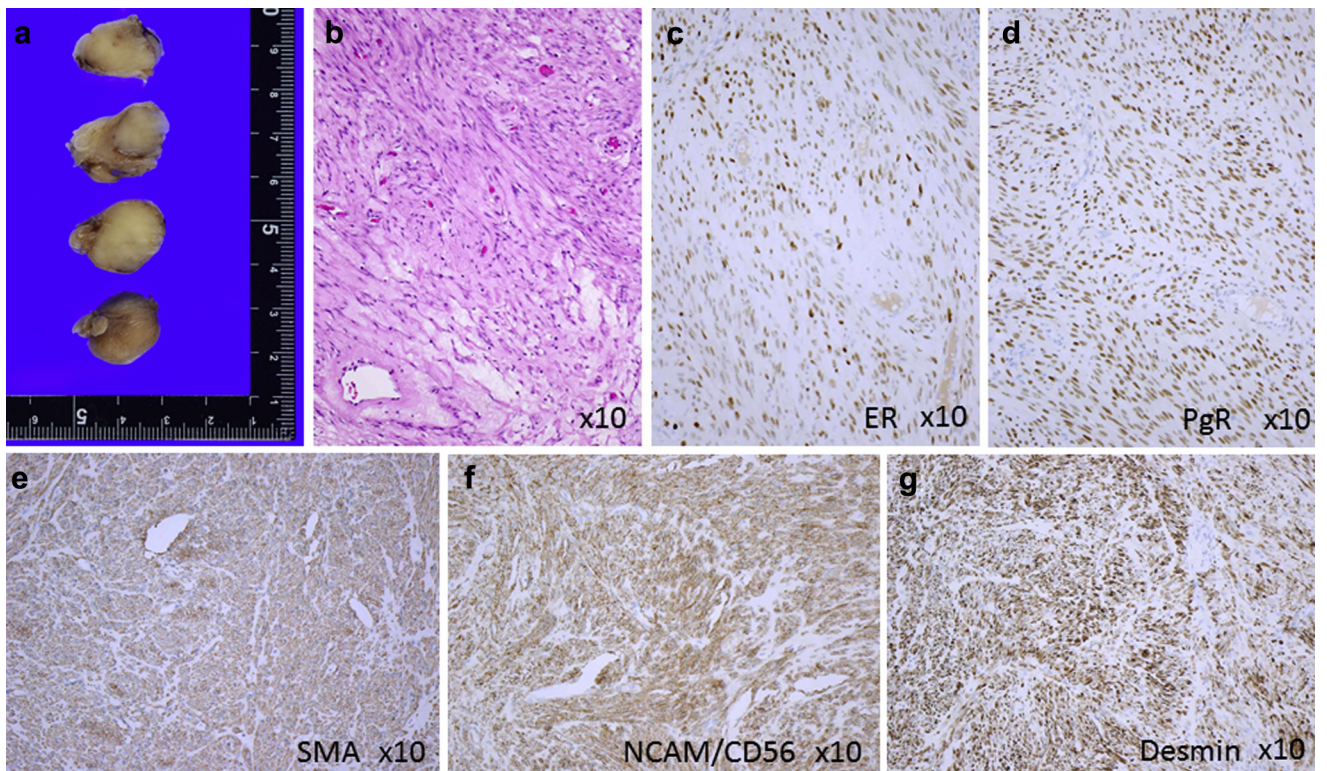


Figure 3. Pathological findings. The resected tumor presents a light grayish sectioned surface with a $35 \times 25 \times 15$ mm in size with multinodular properties. (a) The tumor is myxoid spindle cell tumor which is relatively rich in the small blood vessels (hematoxylin and eosin stain). (b) The tumor has positive hormone receptors (estrogen receptor (ER), progesterone receptor (PgR)), and is diagnosed as leiomyoma, benign smooth muscle neoplasms, rarely encountered in the genitourinary tract outside the uterus. (c,d) The tumor is positive muscular markers (smooth muscle actin (SMA), Desmin) and positive NCAM/CD56 which become positive with myometrium (e–g).

Pathological diagnosis: Urethral angiomatous leiomyoma (Fig. 3)

Pathological findings were an angiomatous leiomyoma which was relatively rich in the small blood vessels with multinodular properties covered with mucous membrane between squamous epithelium and transitional epithelium, and had no malignant findings. The tumor was positive hormone receptors (estrogen receptor (ER), progesterone receptor (PgR)), muscular markers (smooth muscle actin (SMA), Desmin), NCAM/CD56 which become positive with myometrium. The tumor had low cellular density and a cellular atypia. The tumor did not have a mitotic figure and malignant findings in the histology. The tumor was regarded as a recurrence because of the same histological findings as that of 7 years ago.

Discussion

Leiomyoma which belongs to soft tissue tumors, includes deep soft tissue leiomyoma and genital leiomyoma. On the other hand, angioleiomyoma arises from the smooth muscle cells of the venular wall in subcutaneous superficial layer. According to the recent classification of leiomyomas, ER-positive smooth muscle tumors in women (ER-positive SMTs) are considered as a new concept.^{1,2} The tumor in the external urethral orifice in this case has hormone receptors other than muscular markers in angioleiomyomatous histology and was confirmed to be ER-positive SMTs. There are reports of “urethral leiomyoma” and “urethral angioleiomyoma”; however, the urethral leiomyoma needs histopathological consideration related to ER-positive SMTs. ER-positive SMTs occurred in vulva are said to be regarded as an origin of hormone receptor-positive smooth muscle of labia majora and some cases

occur as angioleiomyoma derived from vascular smooth muscle. The present case is regarded as urethral “angiomatous” leiomyoma occurred in subcutaneous superficial layer from the histology.

Urethral leiomyoma is considered to be cured by extirpation of a benign tumor, but the tumor rarely has a relapse.³ This case might be insufficiently extirpated in the first excision, because there was hardly a margin in the first excision and the tumor in the second excision had many nodular lesions. These tumors do not have the differences in cellular density, a cellular atypia, and number of mitotic figures in the histology of the initial and the recurrence tumors, and these tumors have no malignant findings.

Conclusion

Urethral leiomyoma will be necessary to be diagnosed from the concept of new ER-positive SMTs including an examination with related hormone receptors. We reported the present case as urethral angiomatous leiomyoma in the concept of new ER-positive SMTs.

Conflict of interest

None.

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