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Urethral Leiomyoma – Common Pathology, Uncommon Location

Üretra Leiomyomu – Yaygın Patoloji, Yaygın Olmayan Yerleşim

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Abstract

Extra-uterine leiomyomas of urethral origin are rarely encountered neoplasms possessing unique features such as the characteristic growth pattern, diagnostic challenges owing to a long list of possible differential diagnoses, possible cure with surgical management and the unique complications that accompany surgical management. Herein, we report a case of urethral leiomyoma in a middle- aged woman with a brief discussion on the evaluation and management aspects including a concise description of this pathology based on the scarce literature information available.

Keywords: urethra, leiomyoma, case report, extra uterine, mass

Özet

Üretral kökenli ekstra-uterin leiomyomlar, karakteristik büyüme paterni, uzun bir olası ayırıcı tanı listesi nedeniyle tanısal zorluklar, cerrahi tedavi ile olası kür ve cerrahi tedaviye eşlik eden farklı komplikasyonlar gibi benzersiz özelliklere sahip, nadir karşılaşılan neoplazmlardır. Bu yazıda, orta yaşlı bir kadın hastada görülen üretral leiomyom olgusu, değerlendirme ve tedavi yönleri hakkında kısa bir tartışma ile birlikte, mevcut az sayıdaki literatür bilgisine dayanarak bu patolojinin kısa bir tanımı ile birlikte sunulmuştur.

Anahtar kelimeler: üretra, leiomyom, olgu sunumu, ekstra uterin, kitle

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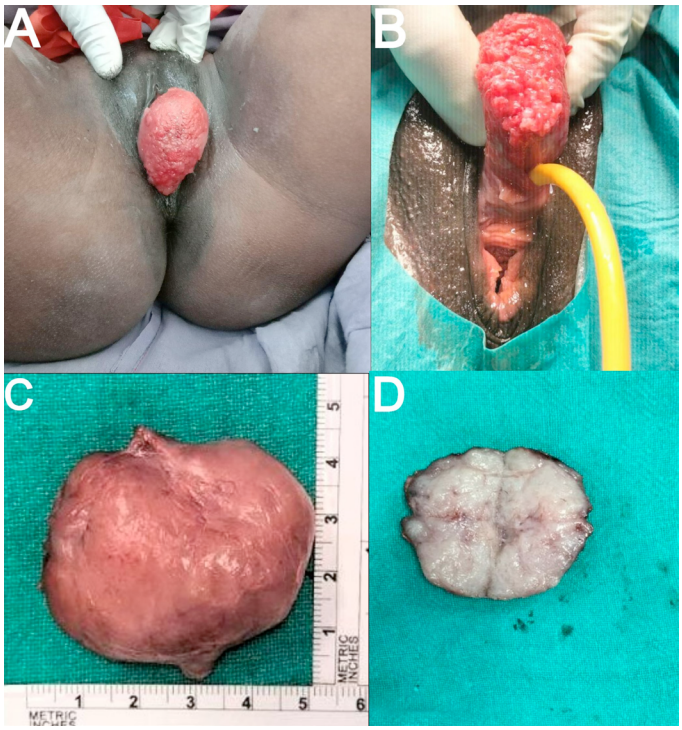

Figure 1. Clinical photographs. A-B: Preoperative photograph; C-D: Specimen photograph

Introduction

As rarely encountered neoplasms, extra-uterine leiomyomas of urethral origin were first reported in 1894 by Buttner et al. [1] They most commonly manifest themselves as perineal masses. Apart from the rarely recognized characteristics of the disease, unique properties such as its characteristic growth pattern and excellent prognosis following surgical excision make it an entity of clinical relevance with good curative possibilities. In addition, the diagnostic difficulties owing to a long list of possible differential diagnoses and specific complications that accompany surgical management make urethral leiomyomas an interesting entity to report with the aim to recognize this pathology, and learn its characteristic features [2].

Case

A 38-year-old female patient without any relevant significant past or family history presented with a progressively enlarging perineal mass for 3 years associated with persistent dysuria and dyspareunia. Examination revealed a 4 x 4 cm nontender, firm and submucosally located perineal mass protruding from the introitus. Focused clinical examination revealed that the mass was arising from the anterior wall of the urethra just proximal to the external urethral meatus **(Figure 1a).** The urethral meatus was pushed inferiorly, and was located at the posteroinferior aspect of the mass **(Figure 1b).** Ultrasonographic examination was suggestive of a 4 x 4 cm mass originating from the anterior urethral region abutting the anterior vaginal wall also showing rich vascularity on colour Doppler ultrasound (US). Pelvic magnetic resonance imaging (MRI) scan revealed the presence of a solid mass lesion originating from the anterior periurethral region, and protruding from the urethral meatus. Fat-suppressed

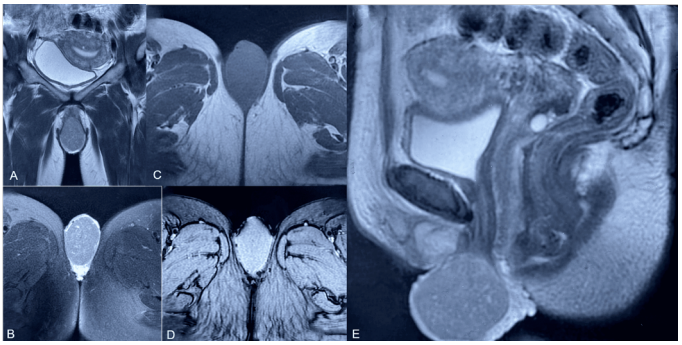

Figure 2. MRI pelvis characterising the urethral leiomyoma. A-E: Coronal & Sagittal T2 images respectively depicting mass in the periurethral region appearing heterogeneously hyperintense

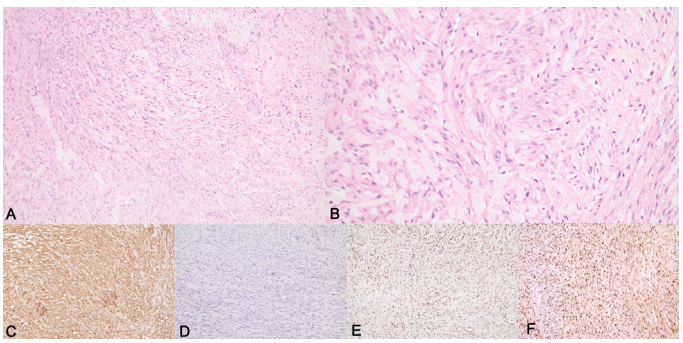

Figure 3. Histological Description. A-B: H & E staining depicting spindle cells with bland nuclei in 100X & 400X magnifications respectively; C: Tumour cells show immunoreactivity for smooth muscle actin (SMA) under 100X magnification; D: Tumour Cells negative for S-100 Immunostaining under 100X magnification; E-F: Tumour cells staining positive for estrogen & progesterone receptor respectively under 100X magnification

T2 -weighted MRI images with intermediate signal intensity did not reveal fat component in the mass, and T1-weighted images were isointense to muscle tissue **(Figure 2a–2e).** A preliminary diagnosis of urethral leiomyoma was arrived at and excision of the mass was planned. Excision of the mass was performed under regional anaesthesia, with preservation of the surrounding tissues excepting the anterior urethral wall. Intraurethral catheter was removed on post- op day 5 and patient urinated normally without any complication. Naked- eye examination of the excised mass revealed a well encapsulated, firm, grey to white coloured mass without any areas of necrosis or haemorrhage **(Figure 1c, 1d).** Histopathological evaluation of the mass revealed the presence of a well circumscribed lesion composed of spindle cells arranged in bundles and fascicles. Cells showed eosinophilic cytoplasm with oval nuclei and no necrosis. Occasional mitotic figures were noted with immunohistochemistry (IHC) that positively stained for smooth muscle actin (SMA) and negatively for S-100, suggesting the presence of a benign leiomyoma. Tumour cells also stained positively for estrogen and progesterone receptors **(Figure 3a–3f).** Patient was asymptomatic at one year follow up with no evidence of any recurrence.

Discussion

Leiomyomas are common encounters in the oncological practice most commonly observed to involve the genitourinary system and notably originating from the uterine musculature. Leiomyomas arising from the urethral region are extremely rare entities. The first case was reported in 1894 by Buttner et al. and up to now only 40 cases have been cited in the literature [1]. Clinically they usually present as slowly enlarging perineal masses associated with complaints like dyspareunia, urethral bleeding or recurrent urinary tract infections. Urethral leiomyomas with similar unique characteristic features seen in only 3 male patients so far, tend to affect women more commonly especially in their 3rd to 4th decades of life. They gradually grow in size during pregnancy and regression noted post-partum suggests a possibility of a hormone- dependent growth potential secondary to expression of oestrogen and progesterone receptors on their surface [2].

Malignant urethral neoplasms are very rarely seen, and usually masses encountered have benign characteristic features. Most commonly observed benign masses are urethral caruncles followed by papillomas and polyps [3]. Needless to reiterate, leiomyomas of urethral origin are extremely rare entities, and most commonly arise from the anterior wall of the proximal urethra [4]. Malignant transformation, and metastases of these benign mass lesions have not been reported so far. Recurrences have been reported in only 2 patients with benign mass lesions treated by repeat excisions [5].

Ultrasound and MRI are the commonly utilised imaging modalities in demonstrating pelvic masses. Especially MRI can be considered the investigation of choice because it provides detailed anatomical description and characteristic signal quantification aiding in accurate histological characterisation. Typical MRI images usually include signal intensities that are isointense to surrounding muscle tissue with signal suppression in fat -saturated sequences and brisk enhancement in post-contrast films [6].

Differential diagnoses among other mass lesions like urethral caruncles, diverticula, polyps, papillomas or haemangiomas should be made. Extremely rare masses of malignant origin include transitional cell carcinoma or squamous cell carcinoma. Surgical excision remains the best treatment alternative for these tumours carrying excellent prognosis owing to very rarely reported recurring potential and unreported malignant transformation [7]. Serious complications following this surgical intervention include urethrovaginal fistula, urethral stricture, stenosis or stress urinary incontinence. Leaving the intraurethral catheter in situ for an extended period of time will help avoid urethral complications [8].

Conclusion

Urethral leiomyomas are extremely rare benign neoplasms with improved postsurgical prognosis following complete surgical excision. Diagnostic dilemmas do exist in the process of establishing the clinical diagnosis of such masses as they are mimicked by tumours of varied histologies predominating the perineal region. Clinicians encountering such cases are requested to keep a keen eye on the evaluation of these urethral mass lesions. Indeed, the combination of local examination, and radiological findings would possibly point towards a definitive and final histological diagnosis of these lesions.

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