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Introduction & Objectives: Primary urethral leiomyoma is a rare benign tumor affecting mostly women aged 30-40 years. Less than 120 cases have been reported in the literature so far. Most frequent location is the posterior wall of the proximal urethra. The clinical presentation often includes symptoms like urinary retention, voiding difficulty, recurrent lower urinary tract infections or dysuria.

Materials & Methods: A 57 year-old woman presented with stress incontinence. Clinical examination revealed a sub-urethral protruding firm tender mass measuring 2.1x1.8x1 cm, which was thought to represent a caruncle. The patient underwent excisional surgery performed under epidural anesthesia. The resection specimen consisted of a well-defined, grey-tan tumor with petechial hemorrhage foci on its surface. On cut section, it showed a grey-white whorled appearance. Histopathology examination described a tumor composed of spindle cells with eosinophilic cytoplasm and elongated blunt-ended nuclei, without significant atypia or mitotic activity, arranged in whorls and fascicles in a hyalinized stroma. Immunohistochemistry tests showed diffuse positive reaction for smooth muscle markers (actin, desmin, caldesmon), negative response for neural tissue, endothelial markers and an inconsequential proliferative index. The final histopathologic report rendered a leiomyoma diagnosis.

Results: A protruding urethral mass can cover up various non-tumor lesions (caruncle, urethral diverticulum, urethrocele, Skene duct cyst) or a neoplastic process arising from the local structures. A rigorous clinical examination, urethroscopy and imaging techniques are mandatory to distinguish between these conditions. However, surgical excision and pathological evaluation confirm the diagnosis.

Conclusions: Urethral leiomyoma is a rare benign neoplasm found in women during their fertility years. Nevertheless, it can occur in the postmenopausal period, as is the current case. The proximal location reflects in the urinary obstructive symptoms, yet in our case the tumor site was atypical and the presentation as well. Tumor resection with a vaginal or abdominal approach, depending on tumor size, is the main curative treatment.