Case Report

Urethral Leiomyoma in Females: A Case Report

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Abstract

Background: Urethral leiomyomas are rare, benign neoplasms arising from smooth muscle tissue that ultimately infiltrate the deep soft tissue of the urinary tract.

Case: A 33-year-old female presented with urinary symptoms and vulvar discomfort due to a palpated growth near the clitoris. MRI revealed a 1.7x2.5x3.1 cm tumor involving the inferior urethra and periurethral tissues. The patient underwent excision of the tumor and cystocopy using general anesthesia. Intraoperative histologic consult after frozen section ruled out overt malignancy and the final diagnosis was leiomyoma.

Conclusion: Leiomyoma frequently occurs in the uterus, but presentation in the urinary tract is exceptionally rare. It is imperative that urethral leiomyomas be pathologically differentiated from their malignant counterparts leiomyosarcomas, which are statistically more common in deep soft tissues.

Introduction

Leiomyomas are benign neoplasms arising from smooth muscle, and are the most commonly diagnosed gynecological tumors, affecting an estimated 70 percent to 80 percent of females [1]. In the clinical setting, leiomyomas commonly present in the subserosal, intramural, and submucosal soft tissue of the uterus and can cause a variety of symptoms including abnormal uterine bleeding, pelvic pain, and bulk or pressure sensations. Although leiomyomas remain asymptomatic for over 50 percent of women, their prevalence and wide breadth of presentations makes them a popular topic of discussion and research within obstetrics and gynecology [2].

In very rare cases, leiomyomas infiltrate the deep soft tissue of the urinary tract. Although more commonly diagnosed in females than males, these benign mesenchymal tumors must be differentiated from their malignant counterparts, leiomyosarcomas. Diagnosis of urethral leiomyoma requires careful physical examination, imaging studies, and ultimately biopsy or surgical resection.

Case

The patient is a healthy 33-year-old female, gravida 2, para 0, abortus 2 who presented for an annual well woman exam complaining of a growth near the clitoris and increased frequency of urination, urinary urgency, and vulvar pain. Pelvic examination revealed the growth as a urethral or periclitoral neoplasm approximately 3 cm in diameter that was rubbery and tender in texture with a flesh-like coloration. All other pelvic anatomy was normal.

Magnetic reasoning imaging was selected as the next study not only to define the extent of the mass, but also to estimate suspicion for malignancy and to optimize the outcome of planned surgical excision. The MRI of the pelvis of the patient revealed a $1.7 \times 2.5 \times 3.1$ cm well marginated, homogenously enhancing solid mass involving the inferior urethra and periurethral tissues that extended caudally to the external urethral meatus and vaginal introiuts. Although the lesion abutted the right crus and posterior body of the clitoris, the radiologist determined the neoplasm arose from the urethra or periurethral tissue.

The imaging appearance was nonspecific and consistent with neoplasm, pending correlation with biopsy or excision. All other female pelvic organs visualized on the MRI were normal. The patient was diagnosed neoplasm of the urethra of low malignant potential and was scheduled for surgical excision of the paraurethral tumor and cystocopy under general anesthesia.

Findings included a 2.0 cm flesh like colored mass, cauliflower like in texture, seen at the external urethral meatus on the perineum, and another 4.0×2.0 cm pink rubbery structure from inside the urethra, almost separated by a thin division.

A 16 F Foley catheter was inserted into the bladder and left in place during the surgery. Marcaine 0.25% with epinephrine was injected into submucosa in the anterior wall of vagina. A 2 cm incision was performed on the anterior vaginal wall under the urethra. The mucosa was divided lengthwise and the external tumor was dissected free. Another tumor from inside the urethral canal became visible at that point. The neoplasm was mobile inside the canal. It was grasped with Babcock clamp and pulled along the urethra and removed in its entirety.

The urethral mucosa was repaired with 3-0 Monocryl on a SH needle in two layers. The vaginal mucosa was repaired in two layers with 3-0 vicryl. The original 16 F Foley catheter was replaced with a larger diameter catheter (20 F) for tamponade and to provide a scaffold for healing. The patient was discharged with a leg bag and the indwelling catheter left in for one week.

After excision was complete, gross examination by the pathologist revealed two tan tissue pieces measuring $1.7 \times 1.0 \times 1.0$ cm and $6.0 \times 1.3 \times 1.0$ cm. Representative sections of both tissue samples were submitted for frozen section examination intraoperatievly. The remainders of the specimens were fixed in formalin for submission. Frozen section examination revealed smooth muscle tumor and polypoid lesion and ruled out overt malignancy.

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Microscopic examination of the formalin-fixed specimens confirmed the final diagnosis of the first specimen as polypoid squamous and urothelial mucosa and the second specimen as a leiomyoma. The leiomyoma was composed of bland spindled smooth muscle cells negative for significant cytologic atypia, mitotic activity or coagulative-type necrosis.

The patient presented six days postoperatively for removal of the Foley catheter inserted during the surgical procedure. Physical examination revealed the perineal and urethral excision site healed well. The patient had no complaints and was able to void without difficulty and did not suffer stress incontinence. The remainder of the patient's postoperative recovery was uneventful.

The urinary symptoms, namely urgency and frequency, and the vulvar discomfort with which she originally presented had resolved.

Discussion

Neoplasms of the urethra are a rare finding and pose a challenge to gynecologists. They encompass a wide breadth of pathology, and it is standard protocol to submit a tissue sample for histologic review. The differential diagnoses of tumors arising from or around the urethral opening include urethral prolapse, urethral caruncle, papilloma, urethral diverticulum, Skene's duct cyst, Gartner's duct cyst, urethral carcinoma, and ectopic ureteroceles [3]. Symptoms reported in previous cases of urethral neoplasms include dyspareunia, repeated urinary tract infections, urinary retention, hematuria, and renal failure [4].

A urethral neoplasm diagnosed as leiomyoma, like in this case, is incredibly rare. The importance of biopsy or compete excision of the urethral mass should not be understated. Tumors of smooth muscle origin, like leiomyomas, infiltrating deep soft tissue like the urinary tract are statistically more likely to be diagnosed as a malignant neoplasm, the leiomyosarcoma.

The urethral leiomyoma affects women more often than men, and most commonly appears during 'reproductive age' from menarche to menopause [5]. It can affect the distal urethra, but the proximal segment is the commonest site, and held true in this case. Urethral leiomyoma has been reported to enlarge during pregnancy and shrink after delivery, suggesting a possible hormonal dependence [6]. Hormonal treatment with a GnRH-agonist is known to cause myoma shrinkage in the range of 34% to 61%. After the cessation of therapy, however, a rapid re-growth of the fibroids often occurs simultaneously with the increasing estrogen levels [7]. Prognosis of urethral leiomyoma is excellent because malignant transformation has not been reported to date and recurrence is rare; however, complete local excision is recognized as treatment of choice as research on hormonal manipulation of urethral myomas is in the preliminary stages.

A particularly fascinating note here is that the MRI described one continuous mass in the urethra, but there were two separated masses ultimately collected during surgical excision, one of which measured at 6 cm in length (twice as long as the single mass described by the MRI). Only after excision of the smaller, oval shaped mass that protruded from the anterior urethra was the second mass discovered and removed. This intraoperative finding upholds recommendation of complete surgical excision.

In the present case, the patient presented for a routine well woman exam and only reported the mass near the clitoris after being asked if she had any abnormal symptoms. Of course, the mass would have been visualized or palpated during the pelvic exam even if the patient had not stated the mass as a current symptom. It is important to note here that the mass had likely been increasing in size for months, and was eventually large enough to cause urinary symptoms and pain.

If pelvic exam of a sexually active female of reproductive age was not standard practice during a preventative well woman exam, the possibility of this particular urethral mass remaining undetected by physicians and unreported by the patient is obvious. Furthermore, as cost to the patient is a paramount inhibiting factor to patient health, if the offering of an annual exam covered entirely by insurance was not standard practice, the possibility of this patient not presenting for physical exam until the mass drastically affected quality of life is even more daunting.

Determination of the best management of urethral masses presents an interesting challenge to gynecologists. Because urethral leiomyoma is so rare and not widely studied, there is no standard sequence of management or practice guidelines. In this case, MRI was selected as the appropriate study to assess the exact location of tumor, depth of tissue infiltration, tissue plane, and presence of features suggestive of malignancy. Due to the rarity of urethral masses in conjunction with the length of time the mass in this case was likely present, complete surgical excision was deemed the appropriate management even before imaging studies were obtained. Fortunately, the pathology of the mass was benign. Should a similar case present to physicians in the future, combined use of the pelvic exam, an indepth imaging study, and ultimately a complete surgical excision is recommended for management of urethral masses based on the findings of this case.

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