

## Muellerianosis of the Urinary Bladder, Endocervicosis Type : A Case Report

This case reports muellerianosis of the urinary bladder, showing glandular lesions made up of endocervical type glands, in a 36-yr-old woman. The patient presented with lower abdominal discomfort and pain on voiding. The patient had undergone two Cesarean sections 5 yr and 3 yr earlier. On a pelvic ultrasonography, a well-circumscribed mass, 2.2×0.8 cm in dimension, was found with luminal polypoid projection in the posterior wall of the urinary bladder. The patient had a transurethral resection of the bladder mass. Histologically, the tumor was composed of irregularly shaped glands lined by endocervical mucous epithelium in the muscularis propria of the urinary bladder. Some glands exhibited cystic dilatation and contained mucinous secretions. The glands elicited no desmoplastic tissue reaction. The intraluminal mucin often contained polymorphonuclear leukocytes. The glands were mostly lined by tall columnar and bland looking mucous cells with mucin secretion. The dilated cells were rarely observed. No endometrial component is noted. It would be appropriate to designate this lesion as "Muellerianosis of the urinary bladder, endocervicosis type". Awareness of the lesion and attention to its typical histologic features should facilitate its crucial distinction from adenocarcinoma.

Key Words: Endocervicosis; Muellerianosis; Bladder, Urinary; Endometriosis

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### INTRODUCTION

A variety of hyperplastic, metaplastic and neoplastic glandular lesions can occur in the urinary bladder. Benign glandular lesions include cystitis glandularis with and without intestinal metaplasia, cystitis cystica, nephrogenic adenoma and urachal remnants. Lesions exhibiting müllerian differentiation, such as endometriosis, have been described in the urinary bladder. In 1992, Philip et al. described six women of reproductive age who had endocervicosis in the urinary bladder, a benign müllerian lesion composed of endocervical type glands, which in some instances, was confused with adenocarcinoma (1). Endocervicosis is much less common than endometriosis or endosalpingiosis, and its apparent predilection for the urinary bladder is enigmatic. In 1996, a publication on this subject was reported by Nazeer et al. with a clinicopathologic study of six cases in which it was described as endocervical type glands in urinary bladder (2). They suggested that this lesion, at least some of the cases, might be of müllerian origin and presented a diagnostic significance to avoid misdiagnosis that resulted in unnecessary or inappropriate treatment. In 1996, muel-

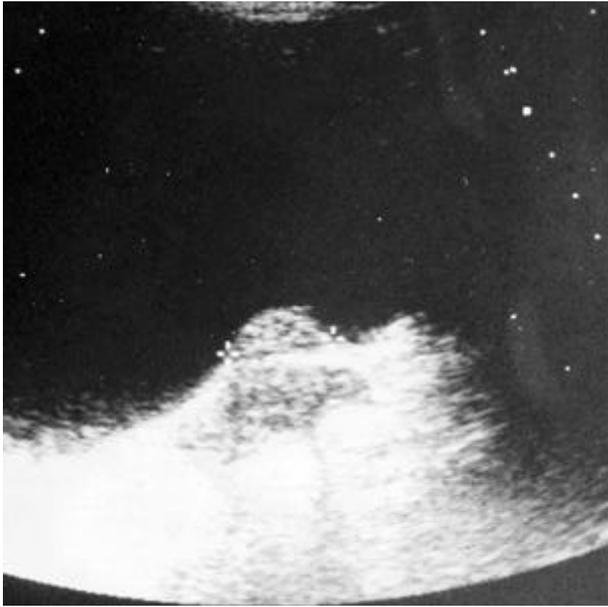
lerianosis of the urinary bladder was designated for such lesions as a distinctive finding in the urinary bladder. Accordingly the lesion should be added to the differential diagnosis of pseudoneoplastic glandular lesions of the urinary bladder (3).

We experienced a case of this benign glandular lesion in the urinary bladder of a 36-yr-old woman. To our knowledge, this is the first case in Korea.

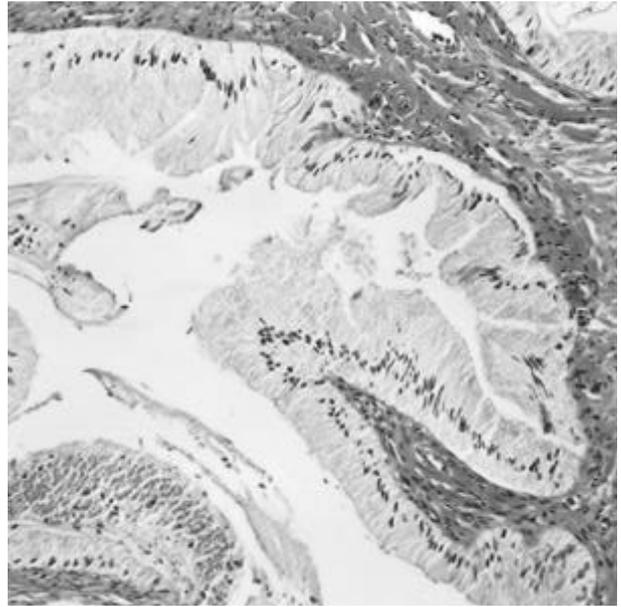
### CASE REPORT

A 36-yr-old woman presented with lower abdominal discomfort and pain on voiding. The patient had a history of gross hematuria with lower abdominal pain for 4 to 5 days in two yr ago, however no clinical treatment was taken.

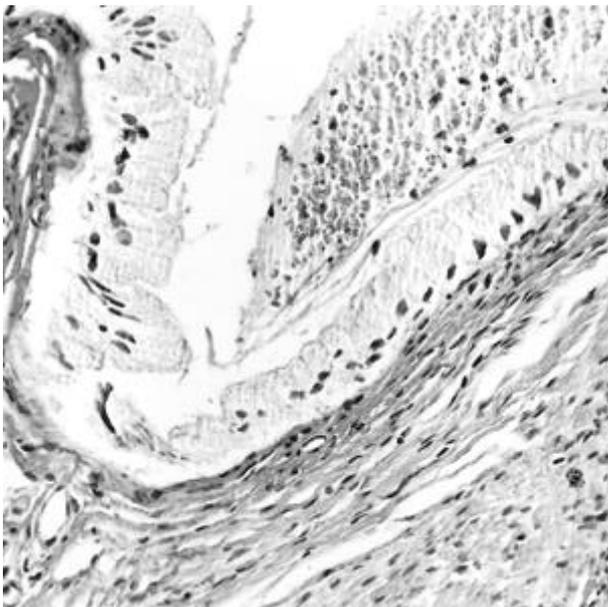
At admission, the routine clinical studies including complete blood count, urinalysis, blood chemistry, simple chest radiography revealed no specific findings. Intravenous pyelography (IVP) study revealed no filling defects in urinary tracts. On a pelvic ultrasonography, a well-defined, polypoid lesion, 2.2×0.8 cm in dimension,



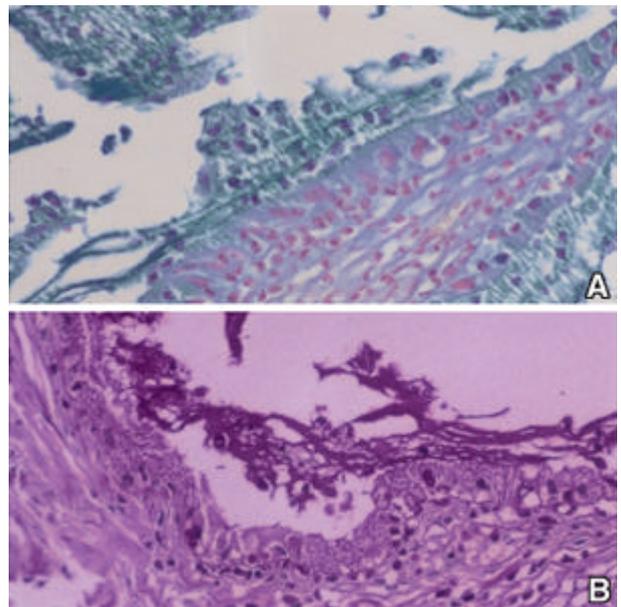
**Fig. 1.** Ultrasonography of the pelvis shows a well-defined, polypoid mass roughly  $2.2 \times 0.8$  cm in dimension in the posterior wall of urinary bladder.



**Fig. 2.** Gland in urinary bladder wall shows mucin containing glands lined by tall columnar mucous epithelium, which is the same epithelium as the endocervical gland of uterus (H&E,  $\times 100$ ).



**Fig. 3.** High power view of the mucous gland shows tall columnar mucin secreting cells with basally oriented nuclei (H&E,  $\times 200$ ).



**Fig. 4.** Alcian blue (A) and periodic acid-Schiff (B) stains show intense staining of both luminal and intracytoplasmic mucinous substances ( $\times 200$ ).

was observed at the posterior wall of the urinary bladder (Fig. 1). The patient underwent cystoscopic examination, which revealed and there was a protruding mass with an intact mucosal surface, suggesting stromal tumor. She underwent the transurethral resection of the tumor.

Grossly, the biopsied specimens were multiple small and irregularly shaped fragments of soft tissue. Histol-

ogically, mixture of variable sized mucous glands reminiscent of the endocervical gland were seen in the urinary bladder wall. The glands were deeply situated within the muscularis propria, but no desmoplastic stromal reaction was noted. The glands were lined by tall columnar mucinous cells with basally oriented nuclei. Some of the glands were cystically dilated and contained intraluminal

mucin with a few polymorphonuclear leukocytes (Fig. 2, 3). Rupture of a gland with extravasation of mucinous contents into the surrounding tissues was also observed. The luminal and intracytoplasmic mucin shows positive reaction in both alcian blue (pH 2.5) (Fig. 4A) and periodic acid-Schiff staining (Fig. 4B). These mucin stains were concordant with those in the uterine endocervix. No endometrial tissue was noted. The nuclei of lining epithelial cells exhibited bland looking appearances. Neither nuclear pleomorphism nor mitose was noted anywhere. The overlying urothelial mucosa was unremarkable except for the mild degree of inflammation.

## DISCUSSION

A wide variety of non-neoplastic lesions can occur in the urinary bladder. Awareness of these lesions is critical to avoid diagnostic misinterpretation. Some of these lesions include von Brunn's nests, cystitis glandularis, cystitis cystica, nephrogenic adenoma and urachal remnants. Misinterpretation of benign lesions as malignant, such as adenocarcinoma, can lead to take unnecessary and inappropriate therapy.

In 1992, Clement and Young reported tumor-like glandular lesions in the urinary bladder of six women, detailing in great depth the clinicopathologic features of these cases (1). The glands showed mucinous, endocervical-type epithelium. The investigators suggested that the lesion was most probably of müllerian origin and proposed the term endocervicosis of the urinary bladder for this entity. They offered the histological similarity to endocervical glands, the location, its occurrence in women, association with endometriosis in some cases, and history of cesarean section in a few cases to support their argument. Ten years earlier, Steele and Byrne reported a case that showed many similarities to the cases described here (4). The authors termed the lesion paramesonephric (müllerian) sinus of the urinary bladder. Recently, Bladamura et al. reported the case of a 52-yr-old woman with a long-standing history of bladder irritative symptoms (5). Transurethral resection of the bladder lesion revealed mucinous glands, including glands resembling endocervical and endometrial glands. This report represents another example of this rare lesion. Awareness of this relatively uncommon lesion is critical to avoid a diagnostic pitfall. The clinicopathologic features including sex, location and histological resemblance to endocervical glands, suggesting that this case might be of müllerian origin. According to the article by Nazeer et al., although the origin of endocervical type glands from the urothelium through metaplasia cannot be completely excluded, the occurrence of this lesion in women only makes this

an unlikely possibility (2).

A differential diagnosis of endocervicosis would include a variety of lesions. The possibility of vesical involvement by minimal deviation adenocarcinoma of the cervix has to be considered (6, 7).

The possibility that this lesion represents a primary, well-differentiated adenocarcinoma of the urinary bladder and secondary bladder involvement by endocervical adenocarcinoma is contradicted by several factors. The lack of an associated endocervical adenocarcinoma, lack of significant cytological atypia and mitoses, desmoplasia and indolent clinical course all support a benign lesion.

Some benign lesions should also be considered in the differential diagnosis. Cystitis cystica and cystitis glandularis can superficially mimic endocervicosis. However, both entities lack the deeply situated glands in muscularis propria seen in endocervicosis.

Urachal remnants occasionally occur in the urinary bladder and should also be considered. They are commonly observed near the apex or anterior aspect of the bladder dome. Histological examination reveals a tubular structure lined by transitional or mucinous epithelium with loose peritubular fibromuscular tissue. But the location and distinctive histopathologic features of this case preclude such a diagnosis.

Nephrogenic adenoma of the urinary bladder is another benign lesion that may be confused with endocervicosis. Although traditionally regarded as a neoplasm, nephrogenic adenoma most likely represents a metaplastic change in the urothelium. Microscopic features of nephrogenic adenoma show small tubules lined by cuboidal and hobnail cells, which are small and lack mucinous lining or deep location in the muscularis propria.

Other entities considered in differential diagnosis are vaginal adenosis and clear cell adenocarcinoma of the vagina in the genital tract of women exposed to diethylstilbesterol in utero. Vaginal adenosis is characterized by endocervical type mucinous glands with frequent squamous metaplasias which are lacking in this case.

In the point of designation of this disease entity, endometriosis with mucinous metaplasia potentially merges with the pathogenesis of endocervicosis in urinary bladder. Examples of endometriosis with mucinous metaplasia have been recorded or illustrated in the urinary bladder (8). But the lesion represents endometriosis admixed with urachal remnants and usually occurs as occasional endocervical-type glands surrounded by endometriotic stroma within an otherwise typical focus of endometriosis. Any focus of endometriosis or endometrial stroma is not found in this case.

In summary, endocervical type gland which represents a benign lesion, can rarely be observed in the urinary bladder. Although the histogenesis of this lesion is not

entirely clear, at least some of the cases may be of müllerian origin. We prefer to designate the diagnostic term of this case as müllerianosis of the urinary bladder, endocervicosis type, because endometrial tissue is not noted in this lesion. Finally recognition of this lesion is important to avoid misdiagnosis that results in unnecessary or inappropriate treatment.

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