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Case report

Mullerianosis of the urinary bladder; A case report

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ABSTRACT

Introduction: Mullerianosis is a sporadic condition of the urinary bladder. The study aims to present a case of mullerianosis with a brief literature review.

Case presentation: A 52-year-old lady presented with hematuria for one-year duration. A general urine examination showed microscopic hematuria. Abdominal and transvaginal ultrasonography revealed a soft tissue mass (13 * 10 mm) within the base of the urinary bladder. Cystoscopy showed 1.0 * 1.3 cm benign-looking mass bulging into the posterior bladder wall covered by a normal bladder mucosa. Transurethral resection was done. The histopathological examination was consistent with mullerianosis.

Discussion: Two hypotheses have been proposed; the first one (implantation theory) believing that at the time of pelvic surgery and caesarian delivery Mullerian tissues become implanted inside the bladder. The second theory (metaplastic theory) holds the probability of differentiation of Mullerian epithelium to endometrial, endocervical, and tubal components and proliferation in the bladder.

Conclusion: mullerianosis is a very infrequent entity that mainly affects the urinary bladder and is mainly reported in females during the reproductive period, medical and surgical treatment are both regarded as the management options.

1. Introduction

Mullerianosis is a sporadic condition that is defined by the presence of an admixture of at least two of three Mullerian-derived components (endosalpinx, endometrium, and endocervix) [1]. It usually appears as a polypoid mass in the area of the lamina and muscularis propria of the urinary bladder [2]. The most frequent combination occurs between endometriosis and endocervicosis, while conjugation of the three types together is very uncommon [2,3]. In the literature, there are only a few cases of this entity, and it was first claimed by Young and Clement in 1996 [4]. Mullerianosis mainly affects the urinary bladder of the women at the reproductive period (from 28 to 53 years) and develops in up to % 50 of patients with a history of pelvic surgery or caesarean delivery [5].

The study aims to present a case of mullerianosis consisting of the combination of all three Mullerian tissue components with a brief review of the literature. The report has been written in line with SCARE 2020 guidelines [6].

1.1. Patient information

A 52-year-old lady referred to urology clinic for having on and off hematuria for one year duration associated with suprapubic pain and dysuria. She was a married ex-smoker (history of smoking was four pack-years), having irregular menstrual cycles with occasional menorrhagia for the last two years. The hematuria was more prominent in the first-morning void, and gradually declined over the day, not necessarily associated with her irregular menstrual cycles. Her past medical history was significant for hyperthyroidism for the last two years (controlled on 5 mgs of methimazole tablets). The past surgical history was significant for 3 previous caesarian sections (the last one being 15 years ago) and ureteroscopy for having a right ureteral stone. Her family history was negative for a similar condition.

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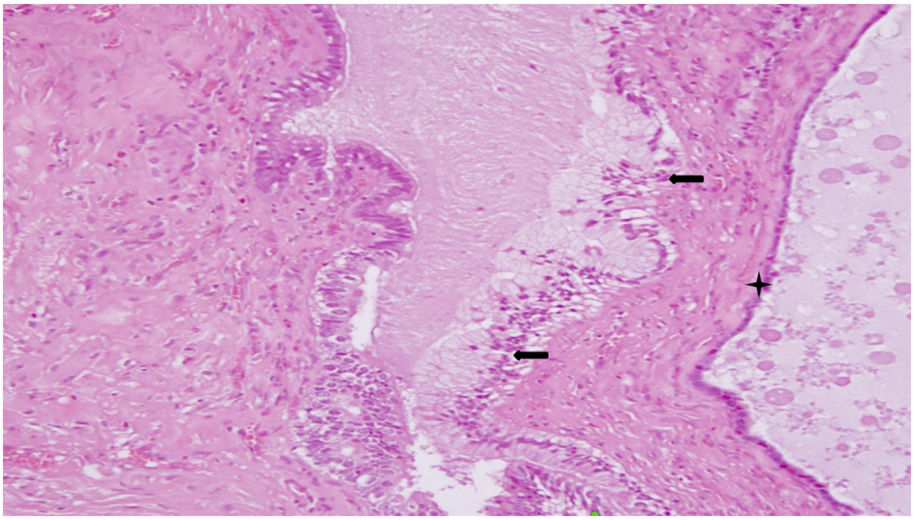

Fig. 1. H&E stain: histopathological image showing the mucinous columnar epithelium, endocervical type (black arrows) and tubal epithelium (black star), in urinary bladder submucosa between the muscular tissue of the bladder wall.

1.2. Clinical findings

There was no significant finding.

1.3. Diagnostic assessment

General urine examination showed microscopic hematuria. Abdominal and transvaginal ultrasonography revealed a soft tissue mass (13 * 10 mm) within the base of the urinary bladder. The rest of the laboratory values were normal, including thyroid function test.

1.4. Therapeutic intervention

Cystoscopy was performed, there was a (1.0 * 1.3 cm) benign-looking mass bulging into the posterior bladder wall, posterior to the trigone, covered by a normal bladder mucosa. Both ureteric orifices and the rest of bladder wall were normal. The mass had a feeling of slight hardness upon resection and no chocolate like material released upon resection. Trans urethral resection of the bladder tumor (TURBT) was done and tissues sent for histopathological examination. The histopathological examination was consistent with mullerianosis (Fig. 1).

1.5. Follow up

The postoperative period was uneventful. The patient was put on a progesterone based drug (VISANNE 2 mgx1) for 3 months. She tolerated the drug very well and clinically improved. The follow up was through outpatient visit.

2. Discussion

In mullerianosis, the ectopic tissue must form conjugationally in the endometrium with at least one of the other tissue types, that is usually seen as a polypoid mass in the bladder wall [2,3]. Regarding the literature, no more than 20 cases have been reported, and very few cases have declared the conjugation of the three Mullerian components together [3,7]. It is worthy to note that, the histopathological examination of the present case revealed the combination of all of the three types together.

The most common affected organ is the urinary bladder, which usually appears in females during the reproductive period, and approximately half of them have undergone pelvic surgery or caesarean delivery [5]. According to Rawan et al. mullerianosis in males has yet to be reported in the literature; however, endometriosis may individually emerge in prostatic cancer males receiving estrogen therapy [2]. Clinical symptoms of mullerianosis vary from dysuria, abdominal pain, hematuria, and recurring renal colic with or without menstruation irregularities [7]. In the present case the patient was a 52-year-old female with a history of hyperthyroidism for two years, complaining of hematuria, dysuria, suprapubic pain and irregular menstrual cycles. The past surgical history was significant for 3 previous caesarian sections.

The decision about this disease must be made carefully Due to the rarity and morphological complexity, which appears as a tumor-like lesion leading to misdiagnosis [2]. Radiographs reveal polypoid mass-like lesion in the posterior wall of the bladder (1 to 4.5 cm) in size [1]. Although, histologically it looks like a glandular epithelial protrusion varying in size in the muscularis and lamina propria of the urinary bladder [2]. Urine cytology is another method that can help to differentiate mullerianosis from other benign and malignant conditions. The occurrence of various sized nuclei within single epithelial cells in a necrotic background increases the possibility of neoplasm while observing macrophages in the urine specimen suggests mullerianosis [8]. Urine endosalpingiosis is very rare while in a study conducted by Jimenez Heffernan et al. endosalpingiosis of the urinary bladder was observed in the urine cytology specimen as such, contained a gross single layer of epithelial aggregates with regular cells, mostly consisted of a small cytoplasm, irregular nuclei and nucleoli [9]. Finding the endometrial cells in a purified urine specimen from menstrual contamination could be a sign of endometriosis or mullerianosis [1]. In the present study, general urine examination showed microscopic hematuria associated with dysuria. Abdominal and transvaginal ultrasonography showed a soft tissue mass.

Regarding the origin of this condition, mainly two hypotheses have been proposed, the first one suggested by Young and Clement (implantation theory), believing that at the time of pelvic surgery and caesarian delivery, mullerian tissues become implanted inside the bladder. The weak point of this theory is that no explanation exists for the presence of mullerianosis in patients with no history of surgical operations or incidence of the condition in other distant body parts [7,10]. Donne et al. proposed metaplastic theory. This theory holds the probability of differentiation of Mullerian epithelium to endometrial, endocervical, and tubal components and proliferation in the bladder. Furthermore, it suggests that because of exclusively locating in the bladder wall peritoneal covering site, it is tremendously responsive to female hormones [11]. In addition, Branca and Barresi argued that the secondary Mullerian system responsible for forming the peritoneal mesothelium might

be able to differentiate into endometrial, endocervical, and tubal tissues [12]. In general, most of the authorities prefer the first theory [2].

A few studies are claiming the transformation of endometriosis to malignancy, mostly adenocarcinoma, with one reported case of adenosarcoma [10]. Mullerianosis could be managed medically and/or surgically. Medical treatment usually comes first except for the condition associated with hydronephrosis. It also includes Estrogen-Progestin contraceptives, progestins, and gonadotropin-releasing hormone agonists [3,13]. In circumstances of non-responding to the medical treatment approximately after 6 months or ureteral involvement, the surgical treatment, including laparoscopic shaving of serosal lesions, partial cystectomy, complete surgical resection will be regarded as the alternative options [3]. TURBT was done for the present case, and follow-up revealed the success of the process.

In conclusion, mullerianosis is a very infrequent entity that mainly affects the urinary bladder and is mostly reported in females during the reproductive period, medical and surgical treatment are both regarded as the management options.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Declaration of competing interest

None to be declared.

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