Article published online: 2023-04-18

Urinary Bladder Mullerianosis Causing Recurrent Abdominal Pain: A Rare Case Report and Review of the Literature

Manjeet Kumar[1] Kailash Chander Barwal[1] Girish Kumar Sharma[1] Kavita Mardi[2] Pamposh Raina[1] Sanjeev Sharma[3]

[1] Department of Urology, Indira Gandhi Medical College & Hospital, Shimla, Himachal Pradesh, India
[2] Department of Pathology, Indira Gandhi Medical College & Hospital, Shimla, Himachal Pradesh, India
[3] Department of Surgery, Rajiv Gandhi Government Post Graduate Ayurvedic College, Paprola, Kangra, Himachal Pradesh, India

Address for correspondence Manjeet Kumar, MBBS, MS, FMAS, MCH, Department of Urology, Indira Gandhi Medical College & Hospital, Shimla, Himachal Pradesh 171001, India (e-mail: manjeetkumar.1014@gmail.com).

J Health Allied Sci[NU] 2024;14:133–136.

Abstract

Mullerianosis of the urinary bladder is a rare bladder lesion with fewer than 30 cases reported in the literature. It describes the ectopic presence of endocervical, endometrium, and endosalpingial tissues inside the urinary bladder. It is diagnosed when at least two of three Mullerian tissues, endometriosis, endocervicosis, and endosalpingiosis, are identified. Mullerianosis presents in females of the reproductive age group, especially after pelvic surgery. Treatment involves resection of the bladder mass and ruling out malignant pathology. Follow-up of Mullerianosis is vital, as recurrence is common. Medical therapy is also indicated when a diagnosis is established after the histopathological examination of the tumor.

We report a 31-year-old female presenting with recurrent lower abdominal pain. Cystoscopy showed a mass of 3 × 3 cm in the urinary bladder. Transurethral resection of bladder tumor was resected, and histopathology suggested a combination of endometriosis, endocervicosis, and endosalpingiosis. Pain was resolved after surgery, but the lesion recurred at 3 months, which was subsequently resected.

Keywords

- Mullerianosis
- urinary bladder
- transurethral resection of bladder

Introduction

Mullerianosis in the urinary bladder is a very rare and complex tumor-like lesion. It is diagnosed when at least two types of Mullerian tissues, that is, endometriosis, endocervicosis, and endosalpingiosis, are seen in the urinary bladder tumor specimen.

Clement and Young first described Mullerianosis in 1996.[1,2] This is seen in young females, especially after pelvic surgeries, typically hysterectomy, caesarean surgery, etc. These lesions present with dysuria, hematuria, and pain lower abdomen. Radiologically, it presents as a mass in the posterior wall or dome of urinary bladder. Mullerianosis of a urinary bladder resembles bladder tumors, so transurethral resection is vital for diagnosis and treatment. We present a case report of Mullerianosis in a female patient presenting with recurrent lower abdominal pain.

Case Summary

A 31-year-old female presented with recurrent pain lower abdomen and dysuria. She did not have a history of hematuria, pyuria, or previous pelvic surgery. She had consulted multiple gynecologists with no relief. On evaluation for lower abdominal pain, detailed history and clinical examination were done. Abdominal examination and per speculum

article published online
April 18, 2023

DOI https://doi.org/10.1055/s-0043-1764360.
ISSN 2582-4287.

© 2023. The Author(s).
This is an open access article published by Thieme under the terms of the Creative Commons Attribution License, permitting unrestricted use, distribution, and reproduction so long as the original work is properly cited. (https://creativecommons.org/licenses/by/4.0/)

Thieme Medical and Scientific Publishers Pvt. Ltd., A-12, 2nd Floor, Sector 2, Noida-201301 UP, India

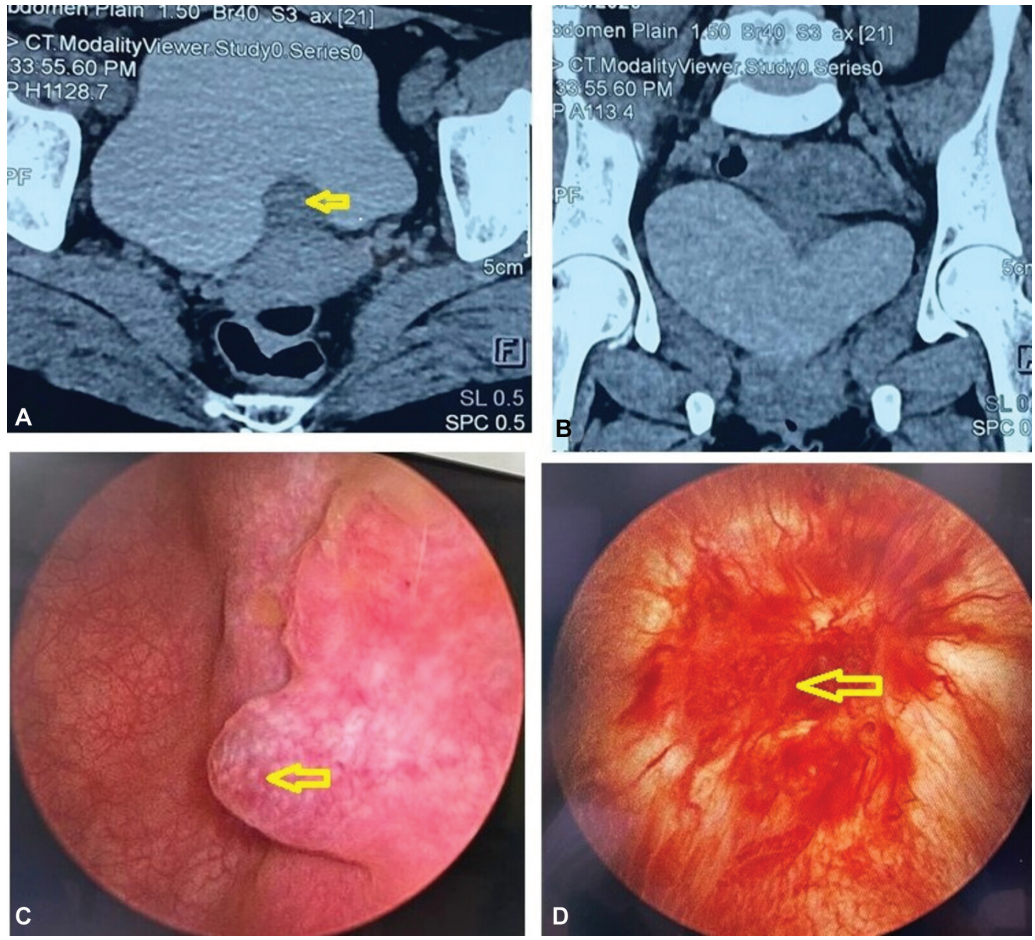

Fig. 1 (**A** and **B**) Contrast-enhanced computed tomography scan showing a mass arising from the posterior wall of urinary bladder. (**C**) Cystoscopy shows a solid mass in the posterior wall of urinary bladder. (**D**) Recurrent lesion at 3 months after transurethral resection of bladder tumor.

examination were normal. Ultrasound abdomen and pelvis suggested a urinary bladder mass for which cystoscopy was planned. On cystoscopy, a mass of approximately 3×3 cm at the posterior wall of urinary bladder was seen. Computed tomography scan pelvis showed a solid mass of 3×3 cm in the posterior wall of the urinary wall. Transurethral resection and biopsy of the mass were done (►**Fig. 1A–D**).

Biopsy revealed an admixture of endometrial glands and stroma along with the endocervical type of glands lined by ciliated cells in the lamina propria and muscularis propria, without cytological atypia or evidence of malignancy in the glands. No urothelial differentiation was noted. The presence of the deeply seated benign glandular structures lined by different types of Mullerian epithelium was consistent with Mullerianosis. All three types of Mullerian tissues—endometriosis, endocervicosis, and endosalpingiosis—were found (►**Fig. 2).**

On follow-up cystoscopy, a residual lesion was seen, which was resected and fulgurated. She was started on a combination of estrogens and progestins. At the 6 months follow-up, no intravesical lesion was seen on ultrasound and cystoscopy.

Discussion

Mullerianosis of the urinary bladder is a rare entity with less than 30 cases described in the literature. It is diagnosed when at least two of three Mullerian tissues like endometriosis, endocervicosis, and endosalpingiosis are identified inside the urinary bladder. It usually affects women of the reproductive age group, that is, from the second to fifth decade of their lives. The symptoms vary from hematuria, dysuria to pelvic pain, which may be associated with menstruation. It most commonly occurs as a polypoid mass in the posterior wall or dome of the urinary bladder. It can mimic a malignant tumor clinically and radiologically; hence, a correct diagnosis can be made only after resection followed by histopathological examination. Histologically, it consists of glands of varying sizes lined by endometrial, endocervical, or tubal epithelium. Mullerianosis is indistinguishable from other tumors that occur in the urinary bladder, clinically or radiologically.[1–3] Habiba et al in a literature review described 27 cases of Mullerianosis in the urinary bladder and three cases in the lower ureter.[4]

In addition to classical endometriosis, glandular lesions made up of endocervical type glands (endocervicosis) can

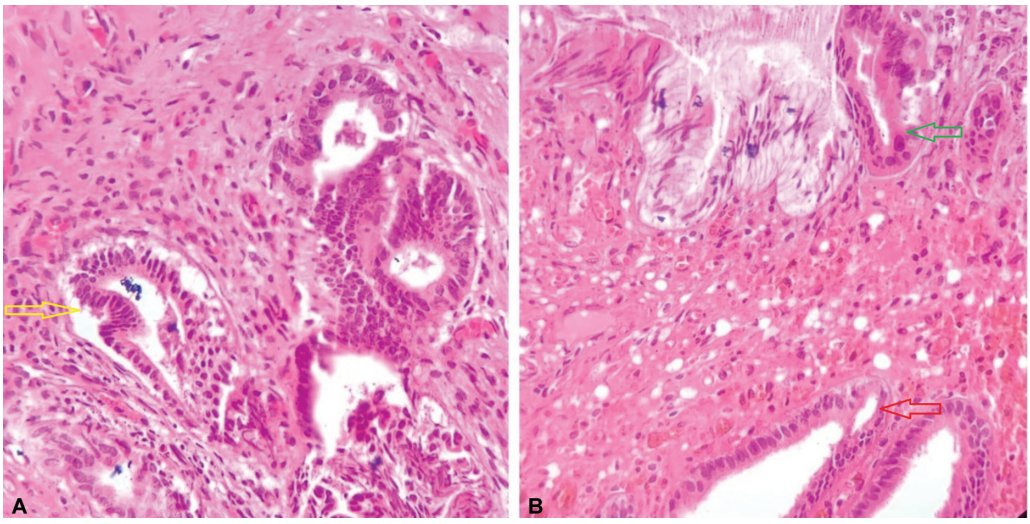

Fig. 2 (**A** and **B**) Histopathology of the resected mass shows endometriosis, endocervicosis, and endosalpingiosis.

also occur in the bladder. Clement and young first described the presence of cervical tissue in the urinary bladder.[1,2] Before this, only endometriosis had been discovered in the urinary bladder. Other Mullerian tissues, such as tubal epithelium in the bladder, were described under the name of endosalpingiosis in case reports.[1,5] The term Mullerianosis is used when there is a combination of any two of the three Mullerian-type lesions.[1]

Two theories have been purposed for the pathogenesis of Mullerianosis. First is the implantation theory, in which Mullerian epithelium gets implanted after pelvic surgeries. However, this theory fails to explain the diagnosis of Mullerianosis in patients with no history of pelvic surgery. Another widely accepted theory is the metaplastic theory, which purposed that the Mullerian epithelium gets differentiated into the endometrial, tubal, and endocervical epithelium. This theory explains the occurrence of Mullerianosis in the posterior wall and dome of the urinary bladder. Peritoneum cells on the urinary bladder get differentiated into Mullerian epithelium on being stimulated by hormones estrogens and progestins.[3,6,7]

Guan et al reported cases of Mullerianosis in females from 28 to 53 years with no history of prior pelvic surgery or caesarean section. He also reported the use of cytology in differentiating it from other bladder tumors.[8]

Endometriosis inside the urinary bladder is seen in females in their second to fifth decades of life, although it can also be seen in postmenopausal females taking estrogen therapy.

Although rarely, endometriosis may also be seen in men with prostatic carcinoma who have received exogenous estrogen therapy. It presents as a mass lesion on cystoscopy in the trigone, posterior wall, and dome of the urinary bladder. About one-third of these cases resemble a solid bladder tumor-like adenocarcinoma.

The differential diagnosis of Mullerianosis includes a variety of lesions both neoplastic and non-neoplastic. Cystitis cystica, cystitis glandularis, urachal remnants, and malignant tumors can mimic Mullerianosis of the urinary bladder. Tubular structures lined by mucinous epithelium suggest urachal remnants; however, they are seen at the dome of the bladder as incidental findings and are usually surrounded by a loose peritubular fibromuscular tissue.[3,9]

Treatment of Mullerianosis consists of medical and surgical options. Medical treatment comprised contraceptive pills containing estrogens and progesterone, progestins, and gonadotropin-releasing hormones.[10] Several case reports reported similar findings and treatment in patients with Mullerianosis.

Urinary bladder Mullerianosis is treated by transurethral resection of the mass and followed up with ultrasound and cystoscopy. Some patients may require medical therapy.

Conclusion

Mullerianosis of the bladder is a rare lesion, which presents as a mass lesion mimicking a malignant tumor. Resection of mass and ruling out malignant bladder tumors is the foremost priority. The possibility of Mullerianosis should always be kept in mind in a young female in the reproductive age group presenting with recurrent pain abdomen and dysuria. Follow-up in the case of Mullerianosis is vital since the recurrence of the disease is common.

Funding
None.

Conflict of Interest
None declared.

Acknowledgments
Thanks to Dr Kalpana Sharma for providing expert advise and technical help.

References

1 Young RH, Clement PB. Müllerianosis of the urinary bladder. Mod Pathol 1996;9(07):731–737
2 Amir RAR, Taheini KM, Sheikh SS. Mullerianosis of the urinary bladder: a case report. Case Rep Oncol 2018;11(01):206–211

3 Kudva R, Hegde P. Mullerianosis of the urinary bladder. Indian J Urol 2012;28(02):206–207
4 Habiba M, Brosens I, Benagiano G. Müllerianosis, endocervicosis, and endosalpingiosis of the urinary tract: a literature review. Reprod Sci 2018;25(12):1607–1618
5 Branca G, Barresi V. Müllerianosis of the urinary bladder: a rare tumorlike lesion. Arch Pathol Lab Med 2014;138(03): 432–436
6 Margulis V, Lemack GE, Molberg K, Saboorian MH. Bladder "Müllerianosis" in a woman with lower urinary tract symptoms and hematuria. J Urol 2001;165(6 Pt 1):1996–1997
7 Donné C, Vidal M, Buttin X, et al. Müllerianosis of the urinary bladder: clinical and immunohistochemical findings. Histopathology 1998;33(03):290–292
8 Guan H, Rosenthal DL, Erozan YS. Mullerianosis of the urinary bladder: report of a case with diagnosis suggested in urine cytology and review of literature. Diagn Cytopathol 2012;40(11):997–1001
9 Nazeer T, Ro JY, Tornos C, Ordonez NG, Ayala AG. Endocervical type glands in urinary bladder: a clinicopathologic study of six cases. Hum Pathol 1996;27(08):816–820
10 Almatrafi MH, Alhazmi AM, Almosaieed BN. Mullerianosis of the urinary bladder. Urol Case Rep 2020;33:101333

 © 2023. The Author(s).