http://www.ejcem.ur.edu.pl/en/ European Journal of Clinical and Experimental Medicine Eur J Clin Exp Med 2019; 17 (3): 277–280

CASUISTIC PAPER

Elżbieta Ostańska © ^{1(ABCGF)}, David Aebisher © ^{2(DFG)}, Ewa Kaznowska © ^{1,3(DFG)}, Dorota Bartusik-Aebisher © ^{4(DFG)}

Bladder Mullerianosis – a case report

¹ Clinical Department of Pathomorphology, Clinical Provincial Hospital No.1, Rzeszów, Poland ² Department of Photomedicine and Physical Chemistry, Faculty of Medicine, University of Rzeszów, Poland ³ Department of Pathomorphology, Faculty of Medicine, University of Rzeszów, Rzeszów, Poland ⁴ Department of Biochemistry and General Chemistry, Faculty of Medicine, University of Rzeszów, Rzeszów, Poland

ABSTRACT

Introduction. Bladder mullerianosis is a rare and proliferative lesion that contains at least two types of ectopic Mullerian tissue in its wall

Aim. To present case of bladder mullerianosis.

Description of case. The text contains a description of a clinical case of a 50-year-old woman admitted to a gynecological ward due to diarrheal symptoms and abdominal pain. In a CT examination of the abdominal cavity with contrast, within the posterior left-sided wall of the bladder a 43x25mm proliferative lesion suggestive of neoplastic character was revealed. Transurethral resection of the lesion (TURB) was performed. Histopathology revealed endosalpingiosis with small endocervical foci. The picture of hyperplasia met the criterion of mullerianosis.

Conclusion. Bladder Mullerianosis is a very rare disease that occurs mainly in women of reproductive age. It has very good prognosis. It is important to differentiate the lesion with malignant tumor. The basis for the diagnosis is the histopathological examination of the lesion tissues taken during the surgery.

Keywords. endocericosis, endosalpingiasis, mullerianosis, urinary bladder

Introduction

Mullerianosis was first described by Young and Clement in 1996 as a rare unit consisting of the endometrium and mucous tissue of the fallopian tube or the cervical mucosa, occurring within the lamina propria mucosa or the muscularis proper to the bladder. Mullerianosis is a mild growth occurring mainly in the posterior wall of the bladder, especially in women of childbearing age. Clinical

symptoms include haematuria, pelvic pain and diarrheal symptoms. ¹⁰⁻¹⁶ From the clinical, cytological and histopathological point of view, this change is similar to tumor growth. A thorough clinical examination, imaging and histopathological examination allow for some differentiation of these hyperplasia. ¹⁷⁻²⁵ Treatment consists of surgical removal of lesions, by transurethral resection (TURB) and, in some cases, homonaglama therapy. ²⁶⁻³³

Corresponding author: Elżbieta Ostańska e-mail: eostanska@wp.pl

Participation of co-authors: A – Author of the concept and objectives of paper; B – collection of data; C – implementation of research; D – elaborate, analysis and interpretation of data; E – statistical analysis; F – preparation of a manuscript; G – working out the literature; H – obtaining funds

Received: 8.04.2019 | Accepted: 10.06.2019 Publication date: September 2019

Ostańska E, Aebisher D, Kaznowska E, Bartusik-Aebisher D. *Bladder Mullerianosis – a case report*. Eur J Clin Exp Med. 2019;17(3):277–280. doi: 10.15584/ejcem.2019.3.14

Description of the case

A 50-year-old woman was admitted to the clinical urology department with diarrheal symptoms and abdominal pain, which initially suggested pyelonephritis. During the distant time, the patient underwent supra-hysterectomy. Urinalysis and urine sediment analysis showed inflammatory changes with the presence of bacteriuria and leucocyturia. In addition, the results of laboratory tests were normal. In computed tomography of the abdominal cavity and pelvis with contrast, a pathological size of 43x25mm was diagnosed within the posterior left-sided wall of the urinary bladder. In the left kidney cup, a 3-mm-thick deposit was revealed, and in the pancreas the focal widened Wirsung cable. In addition, abdominal and pelvic organs without pathological features.

Undergoing general anesthesia, transurethral resection of the bladder was performed. Post-secretion material in the form of numerous tissue fragments was fixed in buffered formalin and tissue and H+E staining was performed. In the microscopic examination, endosalpingiosis was diagnosed with the presence of small endocervikosis foci. The image corresponded to mullerianozie. The image showed fragments of the wall of the urinary bladder covered with mucosa with features of edema. Within it and in the musculature, small, partially cystic glands with lining of the serous type of the fallopian tube and glandular cervix were present, which did not show signs of tumor atypia (Fig. 1).

After about 6 weeks, the patient re-visited the doctor because of abdominal pain. In the ultrasound examination, changes of the infiltrating form in the same

region of the bladder were again diagnosed. The TURB procedure was resumed. In the histopathological examination mullerianosis was again diagnosed. After about 8 weeks after the procedure in the performed magnetic resonance imaging (MRI), in the place after previous surgery, an image suggesting the recurrent nature of the lesions was visualized. In addition, no other pathological changes were observed in the urinary bladder and adjacent organs.

Discussion

Mullerianosis was first described by Young and Clement in 1996 as a rare unit consisting of the endometrium and mucous tissue of the fallopian tube or the cervical mucosa, occurring within the lamina propria mucosa or muscularis proper bladder.1-5 Unlike endometriosis, mulleriasis occurs in the organ, not on the surface of the organ. Other mullerianosis sites are: inguinal lymphatic tubules, ureter, mesosalpinx.1 There are many theories about the pathogenesis of this disease. Theory of implantation, when the changes precede the surgery and the metaplasia theory, when the changes occur in people without a previous operation. There is a suggestion that the Mullerian system, which during the development creates the mesothelial mesothelioma, has the ability to differentiate into the epithelium of the fallopian tube, cervix and endometrium.6 Mullerianosis is a pseudocancer change. The differential diagnosis should include benign and malignant bladder cancer. Clinical symptoms, imaging results, and cytological examination of urine play a role in the diagnosis. The key to the diagnosis is the histopathological examination of change

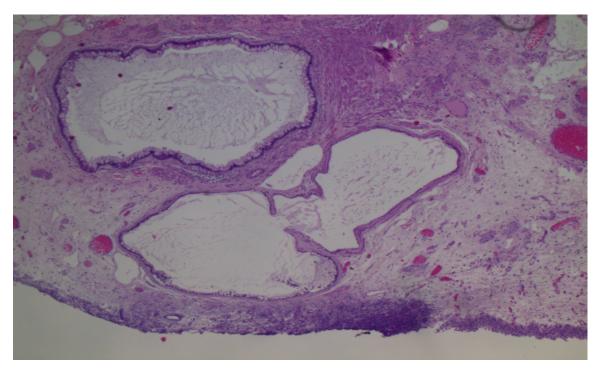


Fig. 1. Bladder Mullerianosis (Staining H+E, area 4X. Own material)

tissues taken during surgery (TURB), which is also an option for the treatment of change.³³⁻³⁵ The second option is conservative treatment with hormone therapy using the LH-RH agonist.⁸⁻¹⁶

Conclussion

Bladder Mullerianosis is a very rare disease that occurs mainly in women of reproductive age. It has very good prognosis. It is important to differentiate the lesion with malignant tumor. The basis for the diagnosis is the histopathological examination of the lesion tissues taken during the surgery.

References

- Yang M, Li Y, Chen M, Chen J, Kung FT. Uterine endosalpingiosis: Case report and review of the literature. *Taiwan J Obstet Gynecol*. 2019;58(3):324-327.
- Petersen RO, Sesterhenn IA, Davis CJ. Urologic Pathology. Lippincott Williams & Wilkins, Wolters Kluwer Health. 2009; 110.
- 3. Bostwick DG, Cheng L. *Urologic Surgical Pathology*. Elsevier. 2014;12.
- 4. Patel A, Desai P, Malczewski F, Stephens D. Mullerianosis of urinary bladder: a rare and problematic bladder tumor. *BMJ Open* 2017; 2017:bcr2016218772.
- 5. Young RH, Clement PB. Mullerianosis of the urinary bladder. *Mod Pathol.* 1996;9(7):731-737.
- Branca G, Barresi V. Mullerianosis of the urinarry bladder: a rare tumorlike lesion. Arch Pathol Lab Med. 2014;138(3):432-436.
- 7. Kudva R, Hegde P. Mullerianosis of the urinary bllader. *Indian J Urol.* 2012;28(2): 206-207.
- 8. Stanimir M, Chiu TuLC, Wese S, Milulescu A, Nemes RN, Bratu OG. Mullerianosis of the uriary bllader: a rare case report and review of the literature. *Rom J Morphol Embryol.* 2016;57(2):849-852.
- 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.
- Maeda K, Kojima F, Ishida F, Iwai M, Kagotani A, Kawauchi A. Mullerianosis and salpingiosis of the urinary bladder: report of two cases with reviev of the literaturę. *Int J Clin Exp Pathol*. 2014;7(7):4408-4414.
- 11. Amir RAR, Taheini KM, Sheikh SS. Mullerianosis of the Urinary Bladder: A Case Report. *Case Rep Oncol.* 2018;11(1):206-201.
- 12. Gilbert N, Guo X, Bauer J, Hennig M, Kümpers C, Merseburger AS. Intravesical salpingiosis: case report and review of the literature. *Aktuelle Urol.* 2018;49(3):266-268.
- 13. Habiba M, Brosens I, Benagiano G. Müllerianosis, Endocervicosis, and Endosalpingiosis of the Urinary Tract: A Literature Review. *Reprod Sci.* 2018;25(12):1607-1618.
- 14. Nakaguro M, Tsuzuki T, Shimada S, Taki T, Tsuchiyama M, Kitamura A, Suzuki Y, Nakano Y, Ono K. Adenocar-

- cinoma arising in urinary bladder endocervicosis. *Pathol Int.* 2016;66(2):108-109.
- 15. Humphrey PA. Endometriosis, endocervicosis and müllerianosis of the bladder. *J Urol.* 2014;192(5):1523-1524.
- 16. Rajakumar C, Vilos GA, Vilos AG, Marks JL, Ettler HC, Pautler SS. Combined transurethral and laparoscopic partial cystectomy and robotically assisted bladder repair for the treatment of bladder endocervicosis: case report and review of the literature. *J Obstet Gynaecol Can*. 2014;36(2):141-145.
- 17. Ozel B, Amezcua C, Ballard C. Endocervicosis of the bladder: a case report. *J Reprod Med*. 2005;50(9):723-726.
- Sener A, Chew BH, Duvdevani M, Brock GB, Vilos GA, Pautler SE. Combined transurethral and laparoscopic partial cystectomy and robot-assisted bladder repair for the treatment of bladder endometrioma. *J Minim Invasive Gy*necol. 2006;13(3):245-248.
- 19. Kudva R, Hegde P. Mullerianosis of the urinary bladder. *Indian J Urol.* 2012;28(2):206-207.
- Olivia Vella JE, Nair N, Ferryman SR, Athavale R, Latthe P, Hirschowitz L. Müllerianosis of the urinary bladder. *Int J Surg Pathol.* 2011;19(4):548-551.
- Cruz Guerra NA, Gómez Raposo MD, Baizán García MJ, Prieto Nogal SB, Juan AG, Sierra MP. Mullerianosis of the urinary bladder: a rare entity. *Arch Esp Urol*. 2009;62(2):150-152.
- 22. Koren J, Mensikova J, Mukensnabl P, Zamecnik M. Mullerianosis of the urinary bladder: report of a case with suggested metaplastic origin. *Virchows Arch.* 2006;449(2):268-271.
- 23. Young RH, Clement PB. Müllerianosis of the urinary bladder. *Mod Pathol*. 1996;9(7):731-7.
- 24. Salada RB, Yong D, Ho CSB, Chong YL. Müllerianosis: A Case Report. *J Endourol Case Rep.* 2019;5(3). doi. org/10.1089/cren.2019.003
- Casasayas-Carles P, Fuentes-Marquez I, Tarrasa-Sagristá F, Gutiérrez Sanz-Gadea C. Müllerianosis of the urinary bladder: report of three new cases. *Arch Esp Urol.* 2014;67(9):771-5.
- 26. Olivia Vella JE, Nair N, Ferryman SR, Athavale R, Latthe P, Hirschowitz L. Müllerianosis of the urinary bladder. *Int J Surg Pathol.* 2011;19(4):548-551.
- 27. Liu C, Perisic D, Samadi D, Nezhat F. Robotic-assisted laparoscopic partial bladder resection for the treatment of infiltrating endometriosis. *J Minim Invasive Gynecol*. 2008;15(6):745-748.
- 28. Li WM, Yang SF, Lin HC, et al. Müllerianosis of ureter: a rare cause of hydronephrosis. *Urology*. 2007;69(6):1208. e9-11.4.
- 29. Koren J, Mensikova J, Mukensnabl P, Zamecnik M. Mullerianosis of the urinary bladder: report of a case with suggested metaplastic origin. Virchows Arch. 2006;449(2):268-267.
- 30. Walid MS, Heaton RL. Laparoscopic partial cystectomy for bladder endometriosis. Walid MS, Heaton RL. *Arch Gynecol Obstet*. 2009;280(1):131-135.

- Casasola Chamorro J, Gutiérrez García S, Fernández Rojo F, Guerreiro González R, De Blas Gómez V, Gallo Rolanía FJ. Bladder endometriosis. Diagnostic and treatment. Actas Urol Esp. 2003;27(5):394-396.
- 32. Redondo P, Idoate M, Corella C. Cutaneous umbilical endosalpingiosis with severe abdominal pain. *J Eur Acad Dermatol Venereol*. 2001;15(2):179-180.
- 33. Preusser S, Diener PA, Schmid HP, Leippold T. Submucosal endocervicosis of the bladder: an ectopic, glandular structure of Müllerian origin. *Scand J Urol Nephrol*. 2008;42(1):88-90.
- 34. Chapron C, Bourret A, Chopin N, Dousset B, Leconte M, Amsellem-Ouazana D, de Ziegler D, Borghese B. Surgery for bladder endometriosis: long-term results and concomitant management of associated posterior deep lesions. *Hum Reprod.* 2010;25(4):884-889.
- 35. Chapron C, Boucher E, Fauconnier A, Vieira M, Dubuisson JB, Vacher-Lavenu MC. Anatomopathological lesions of bladder endometriosis are heterogeneous. *Fertil Steril*. 2002;78(4):740-742.