

Bladder endocervicosis – A rare diagnosis with a silent presentation

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Abstract

Bladder endocervicosis is a benign disease, characterized by the presence of ectopic endocervical tissue in the bladder wall, with few cases described in the literature. A case of bladder endocervicosis in a 35-year-old female, with the previous history of cesarean section, subtotal hysterectomy, and right oophorectomy is reported here. The patient was asymptomatic and the bladder lesion was detected on pelvic ultrasonography. Transurethral bladder resection was performed and the histopathological analysis revealed bladder endocervicosis. 12 months of follow-up have passed without evidence of clinical or radiological recurrence. Clinical presentation, pathological features, and treatment are discussed.

Keywords: Endocervicosis, endosalpingiosis, mullerianosis, urinary bladder

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INTRODUCTION

Bladder endocervicosis is a benign, extremely rare condition, that is defined by the presence of ectopic endocervical tissue in the bladder wall.^[1,2] Usually, it is associated with endometrial or endosalpingiosis ectopic tissue. The simultaneous presence of at least two forms was designated mullerianosis by Young and Clement, in 1996.^[3] The diagnosis of bladder endocervicosis as alone entity is extremely rare, with 40 only cases described in a review by Chea *et al.* Many of the patients reported in the review presented with pelvic and abdominal pain or urinary symptoms, a few with catamenial exacerbation or dyspareunia, and only four were incidental findings. From the forty patients, only ten patients did not have previous pelvic surgery or cesarean section.^[4]


The most commonly affected organs by this disease are urinary bladder, followed by the vagina and uterine cervix.^[2] Clinical manifestation is nonspecific, but suprapubic pain, hematuria, urinary frequency, and urgency are classical symptoms associated with this disease. Dyspareunia and catamenial exacerbation can also be clinical findings in some cases.^[3,4] Lesions are typically located in the posterior bladder wall or at the dome, with dimensions ranging from 2 to 5 cm.^[1,5] Bladder lesions can be detected as echogenic nodules on the ultrasound, or as low signal intensity lesions on T1 weighted images or as high signal intensity lesions on T2-weighted sequences on the magnetic resonance.^[6] Urethrocystoscopy is also an important method for differential diagnosis as most of the lesions appear covered by intact urothelium.^[7]

The authors present a clinical case of this rare clinical entity that presented itself silently and represented a diagnostic challenge.

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CASE REPORT

A 35-years-old female, nonsmoker, was referred to urology consultation due to an incidental finding of a nodular bladder wall lesion on pelvic ultrasonography. She denied the presence of hematuria, irritative or urinary complaints. The patient had a previous history of a cesarean section, 3 years earlier, that was complicated by a severe pelvic infection that culminated in subtotal hysterectomy and right oophorectomy. A suprapubic pelvic ultrasound reevaluation revealed an intramural solid lesion of the right lateral bladder wall with 37 mm. Urethrocystoscopy showed a nodular elevation located in the transition between the dome and the right lateral wall, with no mucosal abnormalities. Pelvic magnetic resonance imaging demonstrated a thickening of the upper bladder wall with 21 mm × 16 mm, without contrast impregnation, in a probable extravascular topography [Figures 1 and 2].

This patient was proposed for transurethral resection of the nodular lesion for pathological diagnosis. The surgery had no complications, and the patient was discharged on the third postoperative day.

Histological analysis revealed the presence of vesical mucosa with mucous glands in muscularis propria and muscularis mucosa layers, some of these glands were cystic. They were coated by endocervical mucinous epithelium and surrounded by their own stroma [Figure 3]. The superficial epithelium had glandular mucinous metaplastic transformation, and the cells were positive for hormonal receptors, closing the final diagnosis of bladder endocervicosis.

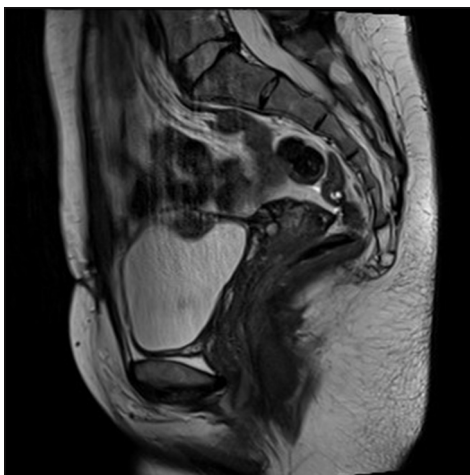


Figure 1: Pelvic magnetic resonance in sagittal T2-weight sequence, showing a hypointense nodule in the bladder dome, with 21 mm maximum diameter and regular limits

At present, after 12 months of surgery, the patient is asymptomatic and without any signs of relapse on pelvic ultrasound.

DISCUSSION

Mullerian lesions derive from the mullerian accessory system and may present as endometriosis, endosalpingiosis, or endocervicosis, with the first being the most common presentation and endocervicosis alone the most exceptional. Bladder endocervicosis is characterized by the presence of endocervical mucinous epithelium in the bladder wall.^[2]

Regarding the etiology, there is a great debate and two major theories. First, the implantation theory, which hypothesizes the migration of endocervical cells by the previous surgery, supported by the fact that most patients have a background of cesarean section or pelvic surgery, as seen in this case report. However, there are cases in which no previous surgery is present going against this theory.^[4,8]

Other authors prefer the metaplastic theory that advocates the transformation of cells of the posterior peritoneum in response to hormonal stimuli. This is supported by the fact that more than one type of mullerian tissue can be present and the lesions are predominantly located in the posterior wall of the bladder covered by the posterior peritoneum.^[2,3,5] The posterior location of the lesion was also a feature seen in the present case.

Given clinical presentation and nonspecific symptoms, differential diagnosis is very difficult because it can be easily misdiagnosed as malignant bladder lesions. Chea *et al.* made a revision of forty cases were noteworthy variability in the symptoms is clear, ranging from hematuria



Figure 2: Pelvic magnetic resonance on coronal T2 fat-suppressed-weight scan, where the same nodule is shown, with right paramedian position

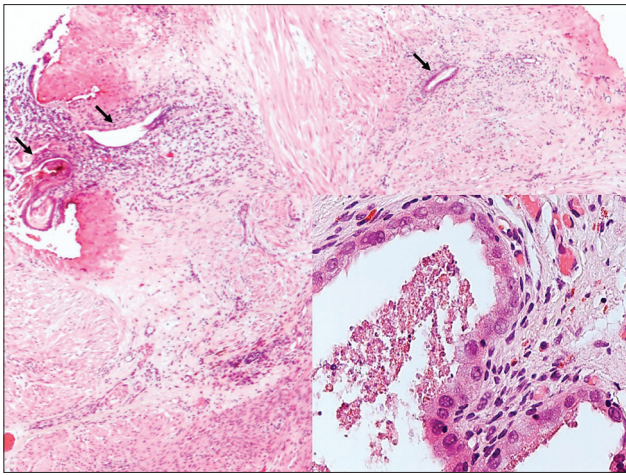


Figure 3: Histology (H and E, $\times 100$): Infiltration of the bladder wall by glands coated by mucinous epithelium (arrows). In the right inferior corner in bigger magnification (H and E, $\times 200$)

to urinary tract symptoms with catamenial exacerbation or dyspareunia, with few patients being asymptomatic like the one presented here.^[4] Urethrocytoscopy findings reveal a mural lesion, generally located on the posterior bladder wall and covered by normal bladder mucosa.^[7]

Differential diagnosis includes cystitis glandularis, cystitis cystica, bladder adenocarcinoma, and carcinoma of the urachus. Definitive diagnosis is established by histopathological analysis, based on cellular architecture and cytological findings.^[2,3,5] Histologically, it is characterized by the presence of endocervical mucin-like epithelium in the bladder wall and may be accompanied by cystic dilatation.^[1] In cystitis glandularis and cystic cystitis, the glands are confined to the muscularis propria layer without extension to the mucosal muscularis layer. In cases of adenocarcinoma, nuclear atypia is present.^[5]

There is no consensus about the best surgical option for managing bladder endocervicosis. The choice depends on different factors, such as the size, location, and number of tumors. Transurethral resection of the lesion is simultaneously diagnostic and therapeutic. There are reported cases in which the therapeutic option was partial cystectomy, either laparoscopic or robotic, with the advantage of complete resection but more comorbidity.^[9] Our approach was transurethral resection, being the patient free of recurrence. Being histopathological analysis benign, it was decided to keep follow-up with regular pelvic ultrasonography.

Despite the benign nature of bladder endocervicosis, a recent case of adenocarcinoma associated to bladder endocervicosis has been reported by Nakaguro *et al.* The

authors have done a review of the literature, describing three other cases of adenocarcinoma originating in endocervicosis from other organs.^[2] These cases, together with the lack of knowledge of the natural course of the disease, point to the importance of keeping these patients under strict surveillance.^[2]

Given the rarity of the disease, there is no standardized follow-up scheme, however, imaging and endoscopic surveillance are recommended, with no definitive time limit.^[5]

CONCLUSION

It is our intention to alert the scientific community to the diversity of bladder tumors, and raise awareness to the importance of the differential diagnosis of bladder disease, where bladder endocervicosis, although rare, should be taken into account.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Humphrey PA. Endometriosis, endocervicosis and müllerianosis of the bladder. *J Urol* 2014;192:1523-4.
- Nakaguro M, Tsuzuki T, Shimada S, Taki T, Tsuchiyama M, Kitamura A, *et al.* Adenocarcinoma arising in urinary bladder endocervicosis. *Pathol Int* 2016;66:108-13.
- Young RH, Clement PB. Müllerianosis of the urinary bladder. *Mod Pathol* 1996;9:731-7.
- Cheah PL, Looi LM, Lee GE, Teoh KH, Mun KS, Nazarina AR. Unusual finding of endocervical-like mucinous epithelium in continuity with urothelium in endocervicosis of the urinary bladder. *Diagn Pathol* 2011;6:56.
- Magill LH, Rajan P, Zafar N, Seywright M, Hendry DS. Endocervicosis and endosalpingiosis of the urinary bladder: A case report. *Br J Med Surg Urol* 2011;4:128-30.
- Eskridge MR, Rovner ES, Payne KD, Workman RB, Curry NS. MRI of endocervicosis: An unusual cause of clustered periurethral cystic masses involving the bladder. *AJR Am J Roentgenol* 2007;188:W147-9.
- McSorley S, Kochman A, DeSouza J. Grand rounds: Müllerianosis of the urinary bladder in a postmenopausal woman. *Urology* 2013;81:488-90.
- Branca G, Barresi V. Müllerianosis of the urinary bladder: A rare tumorlike lesion. *Arch Pathol Lab Med* 2014;138:432-6.
- 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;36:141-5.