

Bladder endocervicosis: Recurrent presentations during pregnancy

Danielle Whiting, Ahmed Ali

Urology Department, Frimley Park Hospital, Frimley, GU167UJ. UK

Abstract

Bladder endocervicosis describes the ectopic presence of endocervical tissue within the bladder. It is a rare condition with an unclear aetiology. We report the case of a 29-year-old woman who presented with bladder endocervicosis during her first pregnancy with recurrence during her second pregnancy. On both occasions, the patient was treated with transurethral resection and the lesion recurred. This is the first case in the literature of bladder endocervicosis presenting during pregnancy and suggests a hormonal stimulus is important in its development.

Keywords: Bladder tumour, endocervicosis, pregnancy

Address for correspondence: Miss. Danielle Whiting, Frimley Park Hospital, Portsmouth Road, Frimley, GU167UJ, UK.

E-mail: danielle.whiting@doctors.org.uk

Received: 13.07.2020, **Accepted:** 11.02.2021, **Published:** 14.07.2021.

INTRODUCTION

Müllerianosis is defined as the ectopic presence of two or more types of Müllerian derived tissue: endometrial, endosalpingiosis, and endocervicosis.^[1] It is a rare condition typically found within the bladder. Solitary bladder endocervicosis is extremely rare with only a few case reports described. It can present a diagnostic challenge mimicking adenocarcinoma.^[2] We describe a case of bladder endocervicosis presenting and recurring during pregnancy.

CASE REPORT

A 29-year-old female presented at 12 weeks into her first pregnancy with abdominal pain and vaginal bleeding. There was no history of hematuria or lower urinary tract symptoms. The patient had no previous abdominal surgery.

A transvaginal ultrasound demonstrated a solid lesion on the right bladder wall. A flexible cystoscopy confirmed the presence of a 2 cm solid lesion near to the right ureteric orifice with a normal appearance of overlying urothelium. Due to its uncertain nature, the patient underwent complete transurethral resection of the lesion under spinal anesthesia at 15 weeks pregnant. The histopathological analysis suggested a diagnosis of the mesonephric remnant.

The patient went on to have a spontaneous vaginal delivery at 37 weeks. During her second pregnancy, an obstetric ultrasound at 13 weeks demonstrated a solid lesion on the right bladder wall. Given the previous finding, a decision was made to await treatment until after the pregnancy. Unfortunately, the patient suffered an intrauterine fetal death at 28 weeks with a subsequent vaginal delivery.

The patient went on to have a further complete transurethral resection of the bladder lesion. Histopathological analysis

Access this article online	
Quick Response Code:	Website: www.urologyannals.com
	DOI: 10.4103/UA.UA_115_20

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How to cite this article: Whiting D, Ali A. Bladder endocervicosis: Recurrent presentations during pregnancy. *Urol Ann* 2021;13:323-5.

was initially suggestive of a mesonephric remnant but due to its unusual appearance and recurrent nature further analysis was performed. This revealed the presence of glands lined by simple columnar mucinous epithelium resembling endocervical epithelium [Figure 1a]. The cells tested positive for estrogen receptor (ER) and progesterone receptor (PR) consistent with a diagnosis of bladder endocervicosis [Figure 1b].

Four months later, magnetic resonance imaging of the pelvis showed a further 2.6 cm mass in close proximity to the right ureteric orifice suspicious for recurrence of her bladder endocervicosis [Figure 2].

The option of partial cystectomy was discussed. As she was planning further pregnancies the patient instead opted for a period of surveillance.

DISCUSSION

Solitary bladder endocervicosis is a rare condition first described as a distinct entity in 1992 with just over 20 reported cases.^[2] As in this case patients are typically women of childbearing age although there are reports in postmenopausal women.^[3,4] To our knowledge, this is the first reported case of a patient presenting during pregnancy.

There are two main theories for the etiology of bladder endocervicosis.^[5] The implantation theory hypothesizes migration of endocervical cells from previous surgery supported by the majority of cases described having previous pelvic surgeries. However, in ours and other cases, this was not the case.^[6] The metaplastic theory suggests the transformation of cells in the posterior peritoneum in response to hormonal stimuli supported by the finding of most lesions on the posterior bladder wall and the expression of HBME-1, ER, and PR.^[7] In our case, the recurrent nature and identification during pregnancy suggest the lesion could have grown in response to a hormonal stimulus. Although, the presentation in

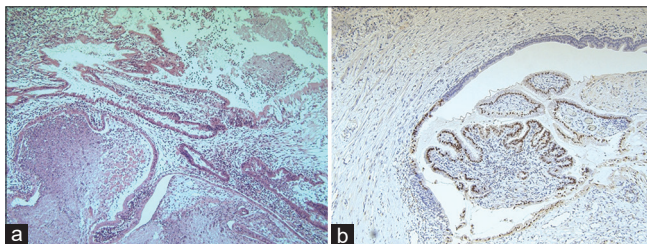


Figure 1: Histopathology from the transurethral resection. (a) Haematoxylin and eosin staining showing the presence of glandular tissue lined by simple columnar epithelium resembling that lining the endocervix. (b) Expression of oestrogen receptor in the endocervicosis glands

postmenopausal women would go against this theory. Bladder endocervicosis is therefore likely to have multi-factorial etiology.

Common symptoms leading to the diagnosis include dysuria, frequency, hematuria, and suprapubic pain. In our case, the patient had lower abdominal pain and vaginal bleeding. However, in the context of pregnancy, it is unlikely these symptoms were related to her bladder endocervicosis. This was more likely an incidental finding as has been described in other cases.^[6,8]

There are numerous differential diagnoses for bladder endocervicosis including adenocarcinoma, carcinoma of the urachus, cystitis cystica, cystitis glandularis, and nephrogenic adenoma.^[2,6] A definitive diagnosis is only reached by histological analysis with the appearance of endocervical-type glands that may be cystically dilated containing mucin.^[9] In addition, immunohistochemical analysis identifying HBME-1, ER, and PR can help support the diagnosis of endocervicosis.^[7]

A variety of different treatment options have been reported including transurethral resection of the lesion and partial cystectomy. In our patient, treatment planning was more complex as she presented during pregnancy. Due to its uncertain nature at the first presentation, we proceeded to transurethral resection during pregnancy after the patient had been consulted on the risks to the fetus. At her second presentation, a decision was made to wait until after pregnancy given she was asymptomatic and the original benign histology. There was no significant progression in the size of the lesion while the procedure was awaited suggesting it is safe to wait until after pregnancy in future cases.

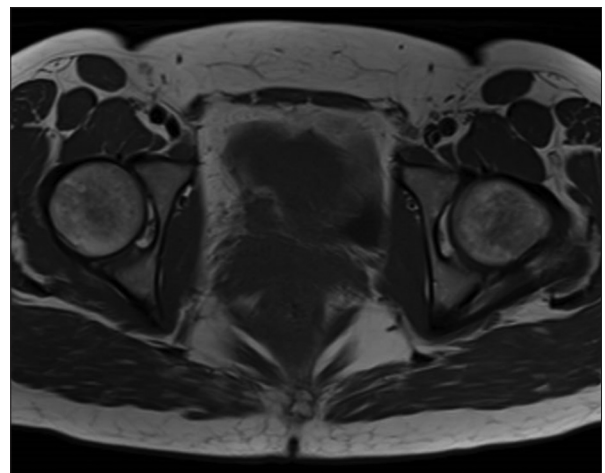


Figure 2: Axial views of the patients T1 weighted magnetic resonance imaging demonstrating a low intensity mass on the right postero-lateral wall

Until recently, bladder endocervicosis was believed to be entirely benign with no malignant potential. However, a case report of adenocarcinoma arising within bladder endocervicosis suggests a need for follow-up.^[10] These lesions are readily visible on ultrasound and cystoscopy. Therefore, the authors would suggest that patients should have annual follow-up after treatment with both modalities. Reporting of patients with this follow-up will then provide a better understanding of the condition and assist in determining follow-up duration.

CONCLUSION

We report a rare case of bladder endocervicosis uniquely presenting and recurring during pregnancy. This implies the role of a hormonal stimulus in its development. Despite the first case being described over 20 years ago the natural progression of the condition remains poorly understood and with the recent description of adenocarcinoma arising within bladder endocervicosis and the evidence of recurrence in our case, these patients must be followed up.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Acknowledgment

We would like to thank Dr. Maria Bahhadi-Hardo

consultant histopathologist for supplying the histology images and Dr. Dhruv Patel consultant radiologist for the imaging.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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