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Case Reports

Adolescent bladder endometriosis initially diagnosed as premenarchal disease: a case report and review of the literature

A. Fujishiro[1], H. Tsubamoto[1], Y. Higuchi[2], A. Kanematsu[2], H. Shibahara[1]

[1] Department of Obstetrics and Gynecology; [2] Department of Urology, Hyogo College of Medicine, Nishinomiya-shi, Hyogo (Japan)

Summary

Only a few cases of premenarcheal endometriosis have been reported in the literature, and bladder endometriosis accounts for 1% of all cases of endometriosis. The authors report the case of an 11-year-old girl who presented with macrohematuria due to bladder endometriosis without apparent menarchal discharge from vagina, and was initially diagnosed as premenarcheal endometriosis. MRI revealed that the lesion penetrated the bladder musculature and infiltrated the anterior wall of the lower uterine segment. She underwent transurethral biopsy and hemostasis, which revealed bladder endometriosis. After three months, she again presented with macrohematuria without exacerbation of bladder endometriosis. Periodic macrohematuria occurred after her basal body temperature dropped. Transrectal ultrasound revealed a fistula in the bladder and the internal cervical os. This is the first report of bladder endometriosis without menarchal discharge, which should be kept in mind when considering differential diagnosis of adolescent macrohematuria without apparent menarche.

Key words: Endometriosis; Bladder; Premenarche; Adolecent.

Introduction

Endometriosis is a chronic gynecologic disorder related to the presence of endometrial glandular and stromal tissue outside of the uterine cavity. It is often associated with dysmenorrhea, dyspareunia, chronic pelvic pain, and infertility. Endometriosis affects approximately 10% of women of reproductive age, which leads to infertility in 30% [1]. It is unusual in adolescents, and only a few cases of endometriosis occurred prior to menarche [2, 3]. Endometriosis of the urinary system is a rare condition, representing 1.2% of all endometriosis cases, of which 84% are in the bladder [4]. The usual site of bladder endometriosis is the dome of the bladder, and 23% of the patients underwent cesarean section [5]. The authors report a case of bladder endometriosis that presented with periodic macrohematuria without menstruation. Informed consent was obtained from the child's parent.

Case Report

An 11-year-old premenarcheal girl presented to a pediatrician because of macroscopic hematuria. She had mild developmental disorder, but her family medical history was unremarkable. Ultrasound examination revealed an elevated lesion, 15 mm in diameter, in the posterior wall of the bladder, and she was referred to a urologist in the present hospital. MRI revealed that the lesion penetrated the bladder musculature and infiltrated the anterior wall of the lower uterine segment (Figure 1), with no abnormality detected in the uterus, vagina, or ovary. In addition, the endocervical gland was developed appropriately for her age, and the authors identified endometrial thickness as physiological change. The preoperative diagnosis was bladder malignancy, with invasion of the anterior wall of the lower uterine segment. Cystoscopy showed that the elevated lesion was continuous to the bladder mucosa in the bladder trigone. No obvious abnormality was noted in either the ureteral orifice or the urethra (Figure 2). Transurethral biopsy and hemostasis were performed. The biopsy specimen revealed bladder endometriosis (Figure 3). The cervix and the vagina were normal. Resistance was felt upon the insertion of the uterine probe into the cervical canal. Three months later, she presented with macrohematuria, but cystoscopy and MRI did not reveal any exacerbation of the bladder endometriosis. Her serum estradiol level was 42 pg/ml, and she presented with macrohematuria after her basal body temperature dropped. However, because of the absence of vaginal bleeding, as in menstruation, transrectal ultrasound was performed. The bladder was invaginated into the internal os, and menstrual blood presumably passed through the fistula between the lower uterine segment and the bladder (Figure 4). The patient has been followed for future repair surgery.

Discussion

Many theories have been proposed to explain the pathophysiology of endometriosis, including retrograde menstruation, metaplasia of the multipotential coelomic epithelium, and embryonic Müllerian rests. Recently, Brosens *et al.* reported that neonatal uterine bleeding

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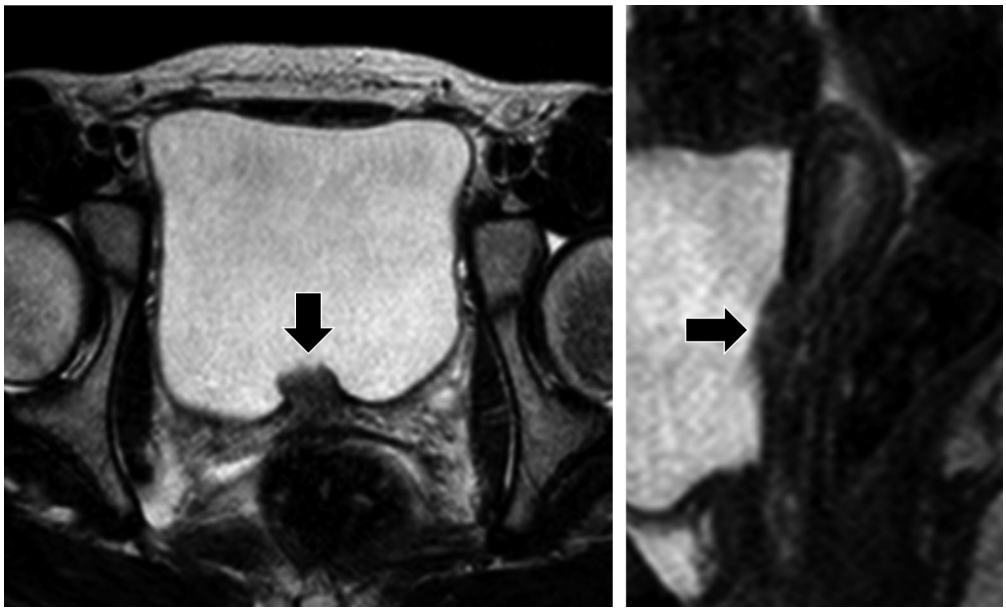

Figure 1. — Magnetic resonance imaging of the intravesical tumor.
(A) Axial, T2-weighted image. (B) Sagittal, T2-weighted image. Arrow indicates a poorly marginated tumor in the lower uterine segment.

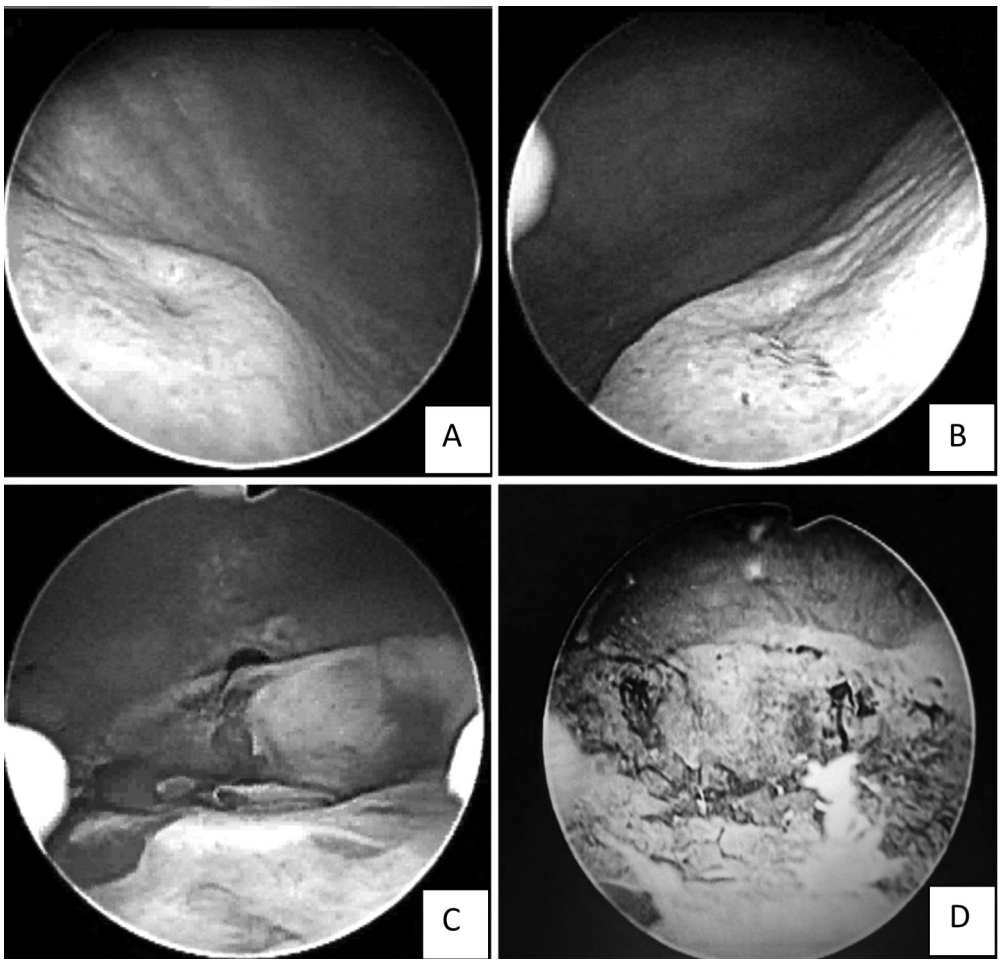

Figure 2. — Cystoscope.
(A) Right orifice. (B) Left orifice. (C) Bleeding from the elevated lesion in the bladder trigone. (D) After biopsy and hemostasis.

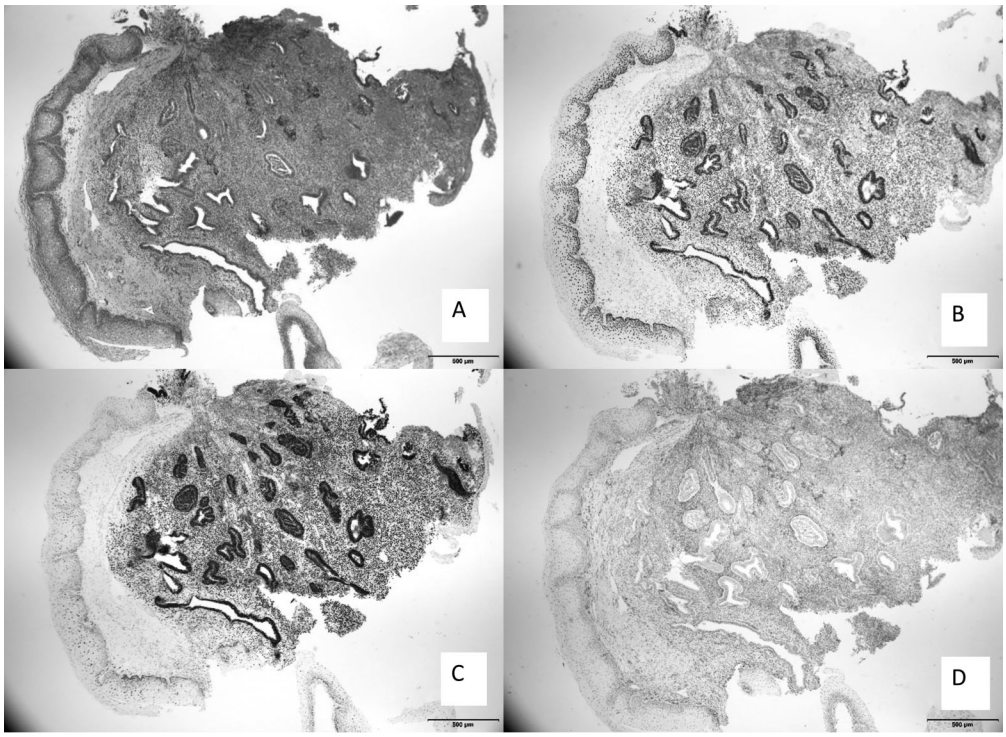

Figure 3. — Pathology of specimen obtained by transurethral resection.
(A) Endometrial glandular cells with squamous metaplasia can be seen under the mucosal epithelium of the bladder (hematoxylin and eosin). Immunohistochemistry shows the endometrial glands stained with both (B) estrogen receptors and (C) progesterone receptors. (D) The endometrial-like stroma is immunopositive for CD10. Bar = 500 nm.

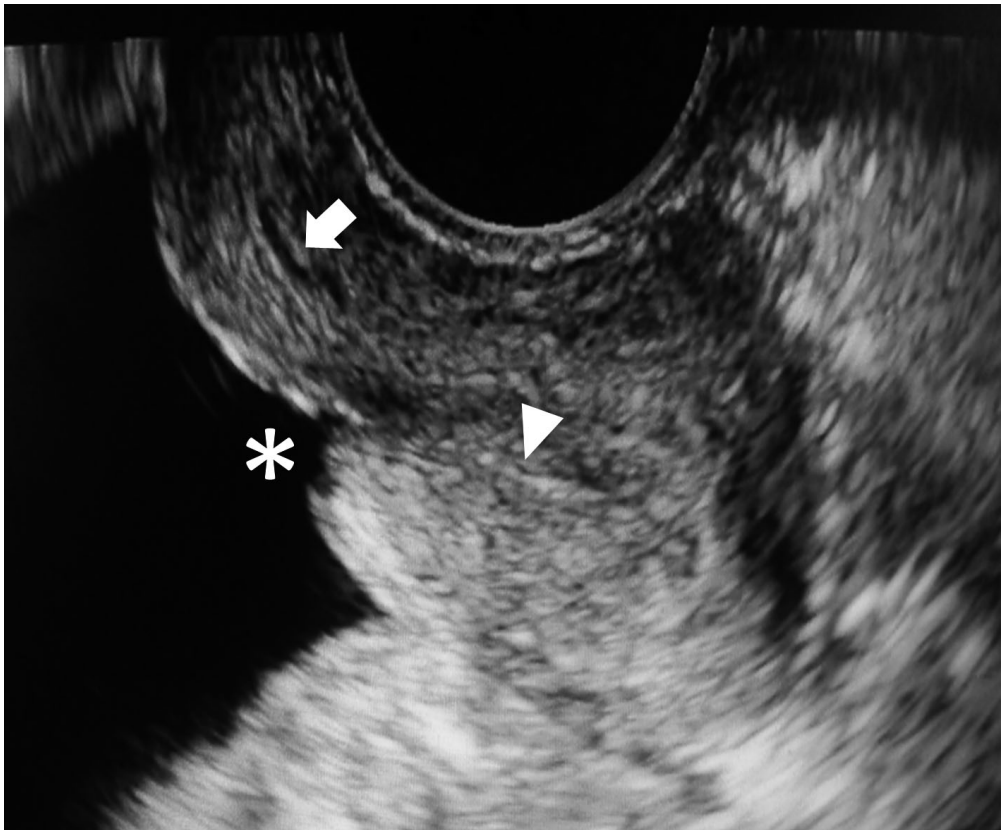

Figure 4. — Fistula between the bladder and the lower uterine segment.
Arrow indicates the cervical canal, arrowhead indicates the endometrium, and asterisk indicates the invaginated bladder toward the lower uterine segment.

caused pelvic endometriosis [6].

The present patient had menses, in the form of macrohematuria, which implicated occlusion of the cervical canal. She might have had congenital cervical dystrophy or congenital vesicouterine fistula. In the literature, however, congenital vesicouterine fistula is rare and generally associated with genital tract abnormalities derived from the Müllerian ducts or urogenital sinus [7, 8].

The use of oral contraceptives pills or progestins to reduce macrohematuria and to prevent exacerbation of bladder endometriosis was considered. The present authors plan to excise the remaining endometriosis lesion and repair the uterus and bladder after the growth of the uterus [9]. The use of OC or progestins was delayed to allow the growth of the uterus.

There are a few case reports of pelvic pain before menarche that were confirmed to be lesions consistent with endometriosis on laparoscopic evaluation [2]. In this case, the girl was referred to the clinic because of macrohematuria; thus, endometriosis must be considered in the differential diagnosis of adolescent macrohematuria.

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Corresponding Author:
A. FUJISHIRO, M.D.
Department of Obstetrics and Gynecology
Hyogo College of Medicine
1-1 Mukogawa-cho, Nishinomiya-shi
Hyogo 663-8501 (Japan)
e-mail: akiko149@hotmail.com